

5-10-2020

New Childhood Chronic Illness: PANS/PANDAS and the Impact on Family Functioning

Jamie L. Dolce

University of Connecticut - Storrs, jamie.dolce@uconn.edu

Follow this and additional works at: https://opencommons.uconn.edu/gs_theses

Recommended Citation

Dolce, Jamie L., "New Childhood Chronic Illness: PANS/PANDAS and the Impact on Family Functioning" (2020). *Master's Theses*. 1471.

https://opencommons.uconn.edu/gs_theses/1471

This work is brought to you for free and open access by the University of Connecticut Graduate School at OpenCommons@UConn. It has been accepted for inclusion in Master's Theses by an authorized administrator of OpenCommons@UConn. For more information, please contact opencommons@uconn.edu.

New Childhood Chronic Illness: PANS/PANDAS and the Impact on Family Functioning

Jamie Lynn Dolce

B.S., University of Connecticut, 2017

A Thesis

Submitted in Partial Fulfillment of the

Requirements for the Degree of

Master of Arts

At the

University of Connecticut

2020

Copyright by
Jamie Lynn Dolce

2020

APPROVAL PAGE

Masters of Arts Thesis

New Childhood Chronic Illness: PANS/PANDAS and the Impact on Family Functioning

Presented by

Jamie Lynn Dolce, B.S.

Major Advisor

Dr. Maria LaRusso

Associate Advisor

Dr. Caitlin Lombardi

Associate Advisor

Dr. Laura Mauldin

University of Connecticut

2020

ACKNOWLEDGEMENTS

To Mom, Dad, and Jillian, thank you for your continued support throughout my years of graduate school! Thank you for listening when I first told you about PANS/PANDAS and my goals for achieving my Master's Degree. You have encouraged me at every stage of this project and I hope I have made you proud. I love you all so much!

To Jeffrey, we began this journey together and I am forever grateful for your love and support. These two years would not have been easy without you by my side. I look forward to our next adventure. I love you to the moon and back!

Thank you to my advisor, Dr. Maria LaRusso, for taking me on as a student and introducing me to this unique and exciting opportunity. This was a major feat in a short time and I am proud of the work we have accomplished!

I am honored and humbled to have been a part of this project and to have gained the knowledge of families living with a child with PANS/PANDAS. Thank you to the families of this study, I hope the future holds understanding, acceptance, and most of all, happiness.

Table of Contents

COPYRIGHT	II
APPROVAL PAGE	III
ACKNOWLEDGEMENTS	IV
ABSTRACT.....	VII
INTRODUCTION.....	1
PANS/PANDAS Characteristics and Symptomology	2
PANS/PANDAS and Child Functioning.....	5
PANS/PANDAS and Family Functioning	6
Childhood Chronic Illness.....	8
Distinct Features of the PANS/PANDAS Experience	12
METHODOLOGY	14
Phase One: Online Parent Forum	14
Phase One: Data Collection.....	16
Phase One: Data Analysis	16
Phase Two: In-person Parent Interviews.....	17
Semi-Structured Interviews	17
Family Illness Narratives.....	19
Phase Two: Data Collection	20
Sampling.....	20
Recruitment	20
Procedures	22
Protection of Human Subjects	23
Participants	23

Phase Two: Data Analysis.....	25
RESULTS	26
Determinants of Family Functioning	27
Child Symptoms and Clinical Course	27
Family Resources and Institutional Barriers	29
Close Relationships	32
Family Sub-Systems.....	32
Parent Relationship (Marriage and Co-parenting).....	33
Parent-Child Relationship.....	36
Sibling Relationship.....	37
Support Network – Extended Family and Friends	38
Routines and Daily Life	39
Family Emotional Well-Being	43
DISCUSSION	47
Limitations	53
Implications.....	55
RERFERENCES.....	58
APPENDICES.....	66
A. Participant Recruitment Flyer	66
B. Institutional Review Board Approval	67
C. Participant Consent Form.....	68
D. Concept Map of Four Themes Presented in Results	72

ABSTRACT

JAMIE L. DOLCE

NEW CHILDHOOD CHRONIC ILLNESS: PANS/PANDAS AND THE IMPACT ON FAMILY FUNCTIONING

MAY 2020

The current study examined parents' reports of change in the parent-child relationship and family functioning following the onset of Pediatric Acute-Onset Neuropsychiatric Syndrome (PANS)/Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections (PANDAS) in their child. Research questions explored 1) how families' day-to-day lives and functioning changed since the onset and 2) what challenges the families faced in raising a child with PANS/PANDAS. Research was conducted in two phases: narrative thematic analyses of online parent forum threads followed by semi-structured interviews with parents. Four primary themes were compiled from analyses of both data sources: Determinants of Family Functioning; Close Relationships; Routines and Daily Life; and Family Emotional Well-Being.

It was found that depending on the state of the child (i.e., well or in flare/exacerbation) the family's level of functioning fluctuated. Also impacting family functioning was lack of support from the medical community, child's school system, and from extended family and friends. The marital relationship was found to be greatly impacted, in addition to strains in the parent-child and sibling relationships. Magnified by the uncertainty that surrounds the experience, parents expressed feelings of caregiver burden which often included a transition out of a job to full-time caregiver. Analyses also revealed frequent use of language indicative of trauma to illustrate the experience. The use of trauma metaphors supports the notion that the

ability to maintain family functioning is not solely a factor of resilience or preparedness and is unlike that of most childhood chronic illnesses.

INTRODUCTION

When discussing family functioning, parents are influenced by personal experience, cultural and religious obligations, political environment and occasionally, peer-reviewed and valid research. They often look to experts to decide what is best for maintaining a healthy family system, seeking advice on routines, parenting styles, and parent-child relationships. What is not emphasized are atypical cases of childhood, including illness or disability, and what to do when the expectations of the parent-child relationship and family unit are not met. Families may feel shame or question their parenting abilities when their child does not develop according to plan (Doron et al., 2018; Dyson & Fewell, 1986; Keller & Honig, 2004). When a child does not progress along an expected trajectory, the nature of the family system and processes that comprise family functioning come under stress (Herzer et al., 2010; McClellan & Cohen, 2007). This can be due to a difficult temperament (Dyson & Fewell, 1986), mental or physical disabilities (Doron et al., 2018) or, for an increasing number of families, the introduction of an unexpected childhood chronic illness.

In recent decades, there has been a rapid increase in “new childhood epidemics.” These include the 4-A Disorders (Asthma, Allergies, Autism, and Attention Deficit Hyperactivity Disorder), as well as childhood eczema, Crohn’s disease, celiac disease, gastrointestinal reflux, irritable bowel syndrome, and migraines, among others (Bock et al., 2008; Perro & Adams, 2017). A relatively new childhood epidemic known as Pediatric Autoimmune Neuropsychiatric Syndrome (PANS)/Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections (PANDAS) is particularly challenging due to the impacts on parent-child relationships as well as alterations to family functioning.

The current study examined parents' reports of change in the parent-child relationship and family functioning following the onset of PANS/PANDAS symptoms in their child. Research questions explored 1) how families' day-to-day lives and functioning changed since the onset and 2) what challenges the families faced in raising a child with PANS/PANDAS. The research was conducted in two phases: 1) narrative thematic analyses of online parent forum threads and 2) semi-structured interviews with parents who have a child diagnosed with PANS/PANDAS. Given the rise in childhood epidemics including relatively new diagnoses like PANS/PANDAS, understanding the impact on family relationships is critical for improving outcomes of affected children and the functioning of the family system.

PANS/PANDAS Characteristics and Symptomology

PANDAS was first described by Susan E. Swedo in 1998 after a subgroup of children at the National Institute of Health were identified as exhibiting a sudden onset of obsessive-compulsive disorder (OCD) particularly after experiencing a Group A Streptococcal infection (Chang et al., 2015; Kurlan & Kaplan, 2004; Melerine & Ledet, 2019; Swedo et al., 1998; Swedo et al., 2004; Swedo et al., 2015; Tona et al., 2017; Williams & Swedo, 2015; Zamzow, 2019). This subgroup was classified as suffering from Pediatric Autoimmune Neuropsychiatric Disorders Associated with Streptococcal Infections (Chang et al., 2015; Melerine & Ledet, 2019; National Institute of Mental Health [NIMH], 2019; Swedo et al., 1998; Wilbur et al., 2019). The criteria for diagnosis of PANDAS includes five characteristics: (1) the presence of OCD and/or tics, (2) pediatric onset (prepubertal), (3) episodic course with abrupt onset and dramatic exacerbations, (4) association with Group A Streptococcal infection, and (5) association with neurological abnormalities such as adventitious movements, motoric hyperactivity or choreiform movements (Chang et al., 2015; Kurlan, 1998; Kurlan & Kaplan, 2004; Melerine & Ledet, 2019;

Murphy et al., 2012; NIMH, 2019; Swedo et al., 1998; Swedo et al., 2001; Swedo et al., 2012; Swedo et al., 2015; Wilbur et al., 2019; Williams & Swedo, 2015; Zamzow, 2019).

In the years following the first description of PANDAS, some children displayed behaviors and symptoms that would fall under all but one of the PANDAS criteria, namely, the association with Group A Streptococcal Infection. Doctors and families alike were perplexed and found it difficult to make an official PANDAS diagnosis with no temporal association to strep (Murphy et al., 2015; Swedo et al., 2012). The term PANS was thus developed and broadly defined as children with sudden onset of OCD and psychiatric symptoms resulting from an infection from a variety of bacterial and environmental agents (Chang et al., 2015; Murphy et al., 2015; Swedo et al., 1998; Swedo et al., 2012; Tona et al., 2017; Wilbur et al., 2019). Criteria for diagnosis of PANS is as follows: (1) abrupt, dramatic onset of obsessive-compulsive disorder or severe restriction of food intake, (2) concurrent presence of additional neuropsychiatric symptoms such as anxiety, behavioral (developmental regression), sensory or motor abnormalities, etc., and (3) symptoms are not better explained by a known neurological or medical disorder (Chang et al., 2015; Melerine & Ledet, 2019; Murphy et al., 2015; Swedo et al., 2012; Tona et al., 2017; Wilbur et al., 2019). See Wilbur et al. (2019) and Swedo et al. (2012) for the diagnostic tables for PANS and PANDAS. Viral illnesses, bacterial infections (e.g., mycoplasma, Lyme Disease) and environmental triggers, such as mold, have been identified as possible triggers for children who are diagnosed with PANS (Chang et al., 2015; Swedo et al., 2012).

Symptoms of PANS and PANDAS include emotional lability, attention difficulties, separation anxiety, rages/aggression and irritability, age-inappropriate behavior, enuresis or urinary frequency, deteriorations in handwriting, and new bedtime fears/rituals (Chang et al.,

2015; Kraybill, 2019; Melerine & Ledet, 2019; Murphy et al., 2012; Murphy et al., 2015; NIMH, 2019; Swedo et al., 1998; Swedo et al., 2001; Swedo et al., 2012; Swedo et al., 2015; Tona et al., 2017; Williams & Swedo, 2015). Swedo et al. (2012) describes that while there is consistency in the acuity of onset of the co-occurring symptoms, there is extensive variability in the severity and types of symptoms that are experienced.

The nature of the neuropsychiatric symptoms prompts consistent misdiagnosis of PANS/PANDAS. Instead, children are thought to be suffering from OCD, ADHD, Autism, Tourette syndrome, ODD or sensory processing disorder, to name a few (Chang et al., 2015; Gilbert & Kurlan, 2009; Kraybill, 2019; Kurlan & Kaplan, 2004; Murphy et al., 2012; Swedo et al., 2004). Another factor that comprises the uniqueness of this illness is its relapsing-remitting nature (Murphy et al., 2015; Swedo et al., 1998; Swedo et al., 2012). After the initial infection and upon other infections, children with PANS/PANDAS experience periods of symptom exacerbations or “flares,” (Swedo et al., 1998). Following these periods and with the use of treatment, children may return to “baseline,” or returned functioning. For example, during exacerbation a child may regress in their handwriting ability only for it to return to an appropriate level after intervention through antibiotics, natural remedies, IVIG (intravenous immunoglobulin), or a combination of the above. As previously stated, the severity and combination of symptoms will vary from child to child and this is also true after each exacerbation (Swedo et al., 2012). Swedo et al. (2012) explains that a particular child’s symptom profile may be dominated by a set of obsessions and compulsions at first, but other symptoms not previously displayed may develop over time. Often, new symptoms will take the place of others that seem to have disappeared or to have been “cured” by a particular treatment.

It is common for patients to experience an “overnight” change within a few days to a few weeks after infection (Kraybill, 2019; NIMH, 2019; Swedo et al., 2001; Swedo et al., 2004; Swedo et al., 2012). The onset of OCD and/or tic behaviors are considered abrupt if the onset reaches maximal impairment in 24-48 hours (Murphy et al., 2015; Swedo et al., 2004; Swedo et al., 2012). In addition, this abrupt onset is in stark contrast to the gradual development of obsessions-compulsions experienced by children with childhood OCD rather than PANS/PANDAS (Murphy et al., 2015; Wilbur et al., 2019). The abrupt onset comprises the first major disruption to family functioning. Notably, some families are able to pinpoint the exact date after which their child’s symptoms began and the obsessive-compulsive and/or tic behaviors took over their daily life (Swedo et al., 2012). Parents feel certain that their child’s behavior and demeanor is noticeably variant from the child’s previous self (Swedo et al., 1998; Swedo et al., 2012). However, parents are not prepared for the stark shift in their child’s behavior and personality (Kraybill, 2019; Murphy et al., 2015; Swedo et al., 1998; Swedo et al., 2012) or the alterations they may need to make to their parenting style.

PANS/PANDAS and Child Functioning

Several studies have documented the impact of PANS/PANDAS on children’s daily lives and functioning beyond the initial onset. For example, Murphy et al. (2015) found that for children with PANS, especially those with comorbid tics, were more likely to experience impairment in a number of areas. These areas, including school performance decline, handwriting deterioration, food restrictions, and lower quality of life, indicate severe impairment in their level of functioning.

Similarly, Tona et al. (2017), examined changes in children’s functioning during PANS/PANDAS exacerbations. Parents were specifically asked about areas of occupation,

mental function, sensory function, and neuromuscular function and the deficits experienced by their child in these areas when in a PANS/PANDAS exacerbation (Tona et al., 2017). This study found that activities involved in daily living such as, “math, handwriting, extracurricular activities, free play, organized sports, community and family social participation, higher level thinking, attention, memory, sequencing, emotional coping, and energy and drive,” (Tona et al., 2017, p. 1) were especially affected when the child was experiencing an exacerbation of PANS/PANDAS. Additionally, parents reported that during a symptom exacerbation, their child often needed assistance to complete tasks that previously did not require help. As a majority of children in the study experienced deterioration in functioning during exacerbation, facets of their lifestyle are increasingly limited.

While the results presented above further our understanding of the challenges that are faced in daily life by children diagnosed with PANS/PANDAS, the extent to which demands are put on the family and the possible changes to family functioning have not been fully investigated.

PANS/PANDAS and Family Functioning

One area of family functioning that has been investigated in families of children with PANS/PANDAS is the impact on the primary caregiver, typically the mother. Previous research has examined the experience of caregiver burden utilizing the Caregiver Burden Inventories (CBI). The CBI, previously used in studies of Alzheimer’s Disease and Rett Syndrome, has been validated for use with families of children with PANS/PANDAS by Frankovich et al. (2015, 2018) and Farmer et al. (2018). In these studies, caregiver burden was defined as the perceived negative effects of caring for a family member. The CBI was used to identify the level at which the family is impacted by the need to perform caregiving duties and is comprised of five categories: time dependency, emotional health, physical health, social relationships, and

development (adult personal growth). The CBI is completed by the parent/caregiver and is scored, with a higher score indicating higher burden and an indication of need for respite care.

It was found in Frankovich et al. (2018) and Farmer et al. (2018) that parents of children with PANS/PANDAS reported higher levels of burden during the child's first flare than caregivers living with individuals with Alzheimer's disease and comparable levels of burden to caregivers of children with Rett Syndrome, a debilitating neurodevelopmental disorder. This comparability indicates the intense stress placed on the family by the introduction of a PANS/PANDAS diagnosis. Additionally, the relapsing-remitting course of PANS/PANDAS creates deep uncertainty which appears to place significant strain on the caregiver (Farmer et al., 2018; Frankovich et al., 2018). Caregiver burden was found to be higher for parents of children who had to switch schools or frequently missed school due to their illness and for the families in which a parent reduced their work hours to manage the child's illness (Farmer et al., 2018).

Frankovich et al. (2018) also found that although CBI scores increased during flares and times in which the child experienced major impairment, caregiver burden tended to decrease over time while the child received care at a PANS/PANDAS clinic (where the study was conducted). The authors theorized that possible explanations for this pattern of decreasing CBI include: clinical treatment for PANS/PANDAS which results in decreased disease severity and/or clinical benefits of compassionate care and support services; burden is particularly high at first clinic visit and naturally decreases over time; the family adapts to child's symptoms over time; and the ability for the child to manage their own care increases as they become older.

One study, Mettica (2018), examined the broader impact of PANS/PANDAS on the family, including how it varied by family demographics and illness factors. Utilizing a parent-report survey posted on PANDASNetwork.org, Mettica (2018) asked families to report on both

children's symptoms and complete ratings on the Impact on Family Scale (IOFS); edited to specify that answers should focus on the time in which it had the greatest impact on the family (Mettica, 2018). Four subscales of the IOFS were also analyzed including, "financial impact, familial-social impact, parental strain/distress, and parental coping," (Mettica, 2018, p. 64). Mettica (2018) found 1) the greater the symptom severity, the greater the impact on the family; 2) being married was associated with decreased parental strain and an increase in parental coping; and 3) when family income was less than \$50,000, the impact of the child's diagnosis with PANS was greater (i.e., higher score on the IOFS financial subscale).

In sum, the existing studies of PANS/PANDAS' impact on the family described above have demonstrated significant caregiver burden, with impacts exacerbated for families with a single parent/guardian, low income, and/or children with greater symptom severity.

Childhood Chronic Illness

Aside from the small number of studies examining family's experiences coping with PANS/PANDAS, there is a larger literature that has examined the general process of adaptation to a child's chronic illness. Studies tend to investigate the impact of chronic illnesses such as asthma, type 1 diabetes, or cancer and have found that family functioning is not greatly affected. While it appears that most families are able to maintain levels of functionality not unlike those of healthy children, the emotional trauma and distress placed on the family is undoubtedly paramount.

In a critical review, McClellan and Cohen (2007) describe that the impact of a child's illness can range from "minimal disruptions to severe distress and functional limitations," (p. 2). The authors detailed the need to unpack the term "family functioning," into component parts such as family cohesion, family adaptability, family conflict, parent-child relationships and

family problem-solving skills (McClellan & Cohen, 2007). McClellan and Cohen (2007) also highlighted the important fact that measures administered to assess family functioning have been built for families *without* a chronically ill child.

The authors compiled studies that included children with cystic fibrosis, juvenile rheumatoid arthritis, type 1 diabetes/juvenile onset diabetes, asthma, hemophilia, and sickle cell disease (McClellan & Cohen, 2007). Mixed results were found with regard to family functioning where families with chronically ill children did not necessarily report compromised functioning compared to healthy controls; however, across all chronic illnesses parents reported high levels of stress, limited time to engage in other activities or work, and increased restrictions on activities due to treatment regimens and routines. McClellan and Cohen (2007) explored possible explanations for the lack of impaired family functioning in the studies reviewed. First, the studies reviewed did not take into account the differing needs of children at various ages and during developmentally important periods. Second, studies did not include if other siblings were in the home and if also chronically ill or healthy. In a similar vein, the studies reviewed did not place emphasis on the illness severity or the amount of time that the child had been ill. For those who have had ample time to come to terms with the diagnosis, the family may have returned to a level of functionality that mimics that of a family not affected by chronic illness (McClellan & Cohen, 2007).

A similar result in regards to time since diagnosis was found by Popp et al. (2014). Parents of children diagnosed with type 1 diabetes or asthma participated in interviews in the home and were administered The Reaction to Diagnosis Interview (RDI) which assesses the parent's reactions to and feelings about their child's diagnosis. From the RDI, parents were then classified as resolved or unresolved. To be "resolved," parents must indicate a change in their

emotions from the initial emotional response to moving forward and appropriately coping with their child's challenges (Popp et al., 2014). These parents indicate that they have shifted from a focus on the trauma of the diagnosis to focus on meeting the child's needs and preparation for the future. In sharp contrast, those who are still stuck in the intense emotional distress and have not developed a coping style are considered "unresolved," (Popp et al., 2014). Parents who are classified as unresolved often become overly concerned with the possible causes for the illness or why their child was affected and not others. Unresolved parents may be unable to reformulate their beliefs about their child to accommodate a new normal. Popp et al. (2014) found that 59% of parents were resolved and 41% were unresolved, similar to percentages reported in previous studies. The effects of parents' unresolved status manifests in increasing conflict and stress in the home as well as a greater likelihood for less healthy communication amongst family members. Of particular interest was the lack of impact of time since diagnosis on classification as resolved or unresolved. The authors indicated this suggests that the meaning that parents attach to their child's diagnosis remains stable across time (Popp et al., 2014).

Parents in the Gannoni and Shute (2010) study described the need to refocus and put attention on the child's needs rather than on their own inner turmoil and upset. Gannoni and Shute (2010) studied families with children who were at least 1-year post-diagnosis with cancer, chronic renal failure, or type 1 diabetes. Parents and children in this study remarked on the inability to continue the families' active participation in activities after the child's diagnosis, similar to the finding of McClellan and Cohen (2007). This included children experiencing more frequent school absences and restricted participation or inability to participate in sport/extracurricular activities. Overall, the "spontaneity," in their lifestyle was greatly altered to the point that families must make additional preparations before activity (Gannoni & Shute,

2010). The necessity of extra preparation and planning was noted to have an effect on healthy siblings in the home. Siblings often had their own social activities disrupted but were also perceived as having greater maturity by parents. Parents across all three illness types were found to have similarities, especially in emotional reactions. The families were reported as “feeling worried, numb and guilty (for not seeking help earlier),” (Gannoni & Shute, 2010, p. 44).

In a metasynthesis completed by Coffey (2006), this continued sense of worry was a theme found in almost half of the 11 studies of childhood chronic illnesses reviewed. In 5 of the 11 studies in which worry was found to be a theme, parents described that the worry affected their everyday life and also projected into the future. More specifically, parents had the worry of who would take care of their child when they are no longer able. The intense worry also prompted parents to be hypervigilant of health issues and changes. Similar to Gannoni and Shute (2010), the parents’ worry also encompassed the effects of the chronic illness on healthy siblings. Families were concerned with the ability to maintain a family life that would not be disrupted for other children in the home despite the challenges. Another major theme was that of uncertainty. Parents spoke of the uncertainty surrounding the management of the illness and lack of guidance by health professionals. It was found that families had experienced instances in which medical professionals are unable or even unwilling to meet the child’s needs. Feelings of being overwhelmed by the responsibility that is required of a chronic illness had long-lasting effects on the parents. Additionally, it was noted that parents may feel the need to take on the large responsibility while simultaneously feeling the urge to withdraw from their own child. In a majority of the articles reviewed, mothers were the primary caregiver and thus, most of the burden fell to them. Such caregiver burden often included the mother leaving their job to provide full time care for their child. However, this tended to encourage social isolation and financial

struggles. As detailed in other studies mentioned here, increased preparation and planning for activities consumes the lives of these families, ultimately leading to a loss of freedom (Coffey, 2006).

Each of the aforementioned studies have introduced the importance of incorporating the parent perspective in examining the impact of a child's diagnosis with a chronic illness. However, there is still a lack of research specifically unpacking the means by which the challenges and uncertainty surrounding PANS/PANDAS affects family functioning. Additionally, it is thought that the unique course of PANS/PANDAS prevents this chronic illness from being categorized as similar to others.

Distinct Features of the PANS/PANDAS Experience

The unique course of PANS/PANDAS (e.g., sudden onset, relapsing/remitting, variable symptom profiles, and the emotional-behavioral issues) may limit how well the existing literature on the impact of chronic childhood illnesses on family functioning can be applied to families with children affected by PANS/PANDAS. Due to the extreme behavioral manifestations and abrupt onset, a family's first experience with PANS/PANDAS is quite alarming and can be difficult to navigate (Chang et al., 2015; Murphy et al., 2015; Williams & Swedo, 2015). Parents are often concerned that their child has suddenly developed a psychiatric disorder and often look to those professionals before other associations are made.

PANS/PANDAS is thus known as a diagnosis of exclusion; all other possible illnesses or disorders must first be ruled out (Chang et al., 2015; Melerine & Ledet, 2019; Williams & Swedo, 2015). However, this process is often delayed as there are no definitive lab tests for PANS/PANDAS, nor is it currently in the Diagnostic and Statistical Manual of Mental Disorders (NIMH, 2019; Zamzow, 2019). Studies have shown that children and families can experience a

lag anywhere from a few months to multiple years between the onset of symptoms and an official PANS/PANDAS diagnosis (Mettica, 2018; Murphy et al., 2012; NIMH, 2019; Swedo et al., 2001; Tona et al., 2017; Williams & Swedo, 2015; Zamzow, 2019). This lag is often the direct effect of few professionals being able to make such a diagnosis. Barriers to this include, some professionals not believing PANS/PANDAS is real (Zamzow, 2019), medical professionals not being trained to notice the signs and symptoms thus assigning other diagnoses with which they are familiar (Kraybill, 2019; Swedo et al., 2012), and the unique symptom profile of each child that requires extensive review of individual medical history (Chang et al., 2015; Swedo et al., 2012). Another barrier includes the various triggers for PANS/PANDAS such as Lyme, mycoplasma, strep, and mold (Murphy et al., 2015; Swedo et al., 1998). Before a diagnosis is made, children are often assigned labels by other family members or teachers such as “bad”, “difficult”, or “not smart” (Kraybill, 2019).

Acquiring the diagnosis however, often does not allow families to feel at ease. Instead, the relapsing-remitting nature has families awaiting the next exacerbation with the knowledge that their child may never return to their previous selves (Murphy et al., 2012; Swedo et al., 1998; Tona et al., 2017). In addition, because of this pattern there is an inability to reach treatment completion in a way that is experienced by those with other diagnoses such as childhood cancer (Swedo et al., 1998; Tona et al., 2017). Unlike other chronic illnesses, the psychiatric symptoms associated with PANS/PANDAS are known to be the cause of swelling and inflammation in the basal ganglia in the brain (Swedo et al., 1998; Williams & Swedo, 2015). This neurological component poses an additional factor for consideration and analysis that is not necessary for chronic conditions such as asthma or type 1 diabetes. It is thought that

such sustained inflammation in the brain and young onset in developmentally critical years can have a lasting impact on the child's potential.

For these reasons and many more, having a child with PANS/PANDAS may be impacting families in ways that differ from families coping with other childhood chronic illnesses. Understanding that there is still a lack of research examining how the challenges and uncertainty surrounding PANS/PANDAS affects family functioning, the current study has the goal of contributing new knowledge that may lead to better understanding and better supports for these children and their families.

METHODOLOGY

Qualitative research methods focus on depth and gaining extensive knowledge on one particular phenomenon. The main purpose is to deeply explore one topic as it relates to a specific population. The intent is not to generalize to other populations, as is the case for quantitative research (Thomas & Magilvy, 2011). With this concept in mind, a qualitative approach served as the appropriate means for evaluating the subjective experiences of parents whose child has PANS/PANDAS. The current study was completed in two phases: 1) narrative thematic analysis of online parent forum threads and 2) semi-structured interviews with parents who have a child diagnosed with PANS/PANDAS. Each phase will be discussed in detail below.

Phase One: Online Parent Forum

Data were collected from ACN latitudes (<https://latitudes.org>), a website of the Association for Comprehensive Neurotherapy, which includes an online forum for parents of children with PANS/PANDAS. Forums operate when a person (member) writes a message to others, called a "post" (Attard & Coulson, 2012, p. 500). Other members then respond and the culmination of posts on a specific topic is known as a "thread" (Attard & Coulson, 2012, p. 500).

The forum is open to the public, but a profile including a username is needed to make a post or comment. Through the use of usernames, members are anonymous and can decide how much of their story they detail. Several members refer to their family using terms such as “DS: dear son,” “DH: dear husband,” and “DD: dear daughter.” In these ways, the members can share personal experiences without concern that they are identifiable by others. The interactions typically consist of a member briefly describing their situation, then asking the other members a question (Knepper & Arrington, 2018). The respondent members are often those who have been dealing with the illness for a longer period of time and offer advice based on their experience. Across the forum, topics such as difficulty finding a doctor to treat PANS/PANDAS or a child’s persistent school refusal are discussed and parents are undoubtedly vulnerable as they look to the other members for reassurance that they are not alone.

Previous studies have successfully utilized online forums to discover patterns and themes in illness narratives (Attard & Coulson, 2012; Jowett, 2015; Knepper & Arrington, 2018). The advantage to using public online discussion forums is threefold: (1) members of the forum have a place to share experiences and knowledge; (2) the forums provide a mass of naturalistic information as conversations take place devoid of the researcher; and (3) the narratives described in the forum capture the meaning of one’s experience, not just facts (Attard & Coulson, 2012; Jowett, 2015; Knepper & Arrington, 2018). During times of emotional distress, and particularly when parents are new to the diagnosis, these online forums serve as a place of support (Attard & Coulson, 2012; Knepper & Arrington, 2018). Advice on symptom management, treatment options and side effects, and ways to adjust both physically and psychologically are in abundance (Attard & Coulson, 2012; Knepper & Arrington, 2018). Particularly unique to the online discussion forums is the spontaneity of discussion (Jowett, 2015). Members are aware that

their audience are others who share in their experiences and thus, speak freely and openly about their challenges or triumphs (Knepper & Arrington, 2018). Their reality is captured in a way that a similar degree of openness is often difficult to obtain by other means (Jowett, 2015; Knepper & Arrington, 2018).

Phase One: Data Collection

Keywords were used to search for relevant threads in the forum and the keywords that generated the most content related to the primary research questions were utilized. For example, some relevant keywords included: family dynamic, normal, parenting, marriage, and lifestyle. The use of the keyword search served as a means of data reduction, as the ACN Latitudes PANS/PANDAS forum contains over 17,000 threads. Threads discussing topics outside of the parent-child relationship and family functioning were not included in the current study. Prior to coding, each relevant thread was copied in its entirety and pasted into a singular Word document with the title of the thread serving as the title of the project file. The files were then uploaded for analysis to NVivo, a qualitative data analysis software. A total of 84 threads were coded and analyzed, with each thread including between 10-50 posts. The threads analyzed in the current study were posted over several years dating from 2009 to 2018.

Phase One: Data Analysis

Analysis of the parent forum threads was conducted using inductive thematic analysis (Riessman, 2005) and were analyzed to examine the range of parent perceived changes in parent-child relationships and other impacts to family functioning. Inductive thematic analysis is considered a bottom-up approach; the codes and themes are derived from the content of the data (Braun & Clarke, 2006; Braun & Clarke, 2012; Riessman, 2005). In this way, it does bear some similarity to a grounded theory approach as the process of coding is undertaken without the

existence of any pre-existing ideas or concepts (Braun & Clarke, 2006; Charmaz, 2006, pp. 42-47).

To begin, chunks of data such as whole lines or segments were coded (Charmaz, 2006, pp. 47-49). The initial chunks focused on points in the individual posts when family functioning was discussed. Sample codes that were developed from the inductive coding process included: uncertainty, caregiver burden, lifestyle changes, revised or altered expectations, and family disconnect. Descriptions were created in NVivo to ensure that codes were clear and properly applied. After such pieces were separated from the larger amount of text, focused coding was used to analyze the impacts on family functioning. To complete the focused coding, research meetings were held frequently and it was decided which initial codes were significant and required further attention (Charmaz, 2001, pp. 344-346; Charmaz, 2006, pp. 57-59). Data was then analyzed using the line-by-line technique in order to capture the most salient aspects of the families' subjective experience in their own terms (Charmaz, 2001; pp. 341-344; Charmaz, 2006, pp. 50-53). At this point, more descriptive codes were applied to signal phrases or descriptions that serve as examples and testimony to the experience. As a final phase, themes were identified for later review in conjunction with themes from the in-depth interviews.

Phase Two: In-person Parent Interviews

Semi-Structured Interviews. The interviews were incorporated as a second phase with the expectation that the interviews would provide an opportunity to obtain more in-depth and comprehensive narratives capturing the experiences of parents whose children have PANS/PANDAS including, the full story of their journey, the ups and downs in family functioning over time, and their reflections after looking back and recounting the experience in detail. As Murthy (2008) describes, the combination of physical and online data sources

produces robust information on the dynamics of particular groups such as the one studied here. Thus, the interviews were expected to generate insights and perspectives from the parents that might not be captured in the online forum.

Semi-structured interviews were chosen for the current study due to the various advantages and emotional nature of the research topic. Semi-structured interviews are a form of data collection in which the interviewer does not follow a strict list of pre-formed questions. Instead, questions are open-ended and flow from the conversation based on the responses of the interviewee. In this way, meaningful questions are woven into regular conversation which is advantageous in that it often mimics casual and friendly conversation rather than survey-style or formal interrogation (Spradley, 1979). Another advantage to semi-structured interviews is the opportunity for participants to talk about what is important to them and how they interpret the questions posed. The term, “in-depth,” refers to the fact that the respondents hold the knowledge and will be encouraged to elaborate when appropriate so as to convey that the interviewer is a student, eager to learn (Weiss, 1995, pp. 65-71; Spradley, 1979). Through the in-depth interviews, the aim was to holistically understand the processes that are involved in caring for a child with this distinct chronic illness and the changes that have occurred over time within the family system (Thomas & Magilvy, 2011). During each interview, actual descriptions and meanings based on the parents’ lived experiences were provided with less room for assumption and interpretations (Emerson, 2001, p. 28-30). The interviews were able to provide information relating to how family relationships are influenced by a child’s diagnosis in addition to supplying multiple accounts of how a family experiences PANS/PANDAS firsthand.

Interview questions were informed by the coding of the online parent forum data and included topics such as: the illness experience, access to adequate care, and addressing

developmental disruptions. Specific questions that addressed family functioning included: How has PANS impacted your family as a whole? How were your everyday lives and daily activities/routines impacted? How did relationships change (parent/guardian's relationship with child, with other children in the family, with spouse or partner, and relationship among siblings)? With extended family? Friends (both child's and parents')? and How is your family doing now? It should be mentioned that many of these questions were answered spontaneously during the course of describing the full timeline of the family's experience.

Family Illness Narratives. The timeline was established by utilizing "family illness journeys" as a main data gathering technique. "Family illness journeys" represent an adaptation of "illness narratives" (Kleinman, 1988) where personal experience and meaning making around the illness is merged with a detailed exploration of a) the family's experience and b) the child's development (how the child's condition emerged, changed, and came to be understood over time). Utilizing "family illness journeys" as the main data gathering technique, the interviews began by inviting parents to recreate how the history of the child's condition evolved; describing progression of symptoms, steps taken to obtain a diagnosis, treatment and recovery process, relapses or flares, and any ongoing symptom management.

Prior research has indicated the importance of incorporating illness narratives in the study of a "disrupted experience," such as a chronic illness (Bury, 2001, p. 264). Researchers collect the stories of those affected, then create conceptual groupings from the data (Riessman, 2005). As more narratives come together (through the addition of more interviews, for example) a typology is often organized and categorized by theme (Riessman, 2005). This typology is considered to help explain reactions to, management of, and normalization of the illness (Bury, 2001). Further, the way in which each narrative is constructed and events connected in meaning

is determined solely by the “teller” (Riessman, 2005). As time passes, it is increasingly difficult for the teller to be devoid of interpretation of the past. Instead, as described by Riessman (2005), “the ‘truths’ of narrative accounts are not in their faithful representations of a past world, but in the shifting connections they forge among past, present, and future,” (p. 6). It is known that each child with PANS/PANDAS presents differently in their symptom profile and level of symptom severity, contributing to variability across families. For these reasons, the use of illness narratives in the current study grounded the intricate and complex experiences into specific elements that are meaningful in the larger population of PANS/PANDAS families.

Phase Two: Data Collection

Sampling. The sampling approach was theory-based and purposeful with a total of 20 cases including both boys and girls and a range of ages of onset of PANS/PANDAS (from age 2 to 12), with particular attention to maximizing heterogeneity and variation (Patton, 2002). Purposive sampling was necessary as participants were required to have specific characteristics such as being a parent or guardian to a child with PANS/PANDAS (Guest et al., 2006; Hennink et al., 2017). In addition, participants were recruited from Connecticut and surrounding areas as these were within a reasonable distance for researchers to travel to complete an in-person interview. We also used an emerging sampling approach (Patton, 2002), which allowed the researchers to augment the initial sample with additional families to ensure sufficient variability in terms of promptness of diagnosis, symptom severity, overlap with other co-existing disorders, and socioeconomic backgrounds of the families.

Recruitment. Participants were recruited primarily through a flier posted in a local PANS/PANDAS Facebook group (NY/NJ/CT PANS/PANDAS Parents Closed Group) by a parent who is an active member and had participated in a pilot interview. This Facebook group

was considered the ideal context for recruitment because 1) closed Facebook groups are widely used by parents of children with PANS/PANDAS (a quick search yields regional groups with thousands of members for almost every part of the United States and several countries throughout the world), 2) participants are from the geographic area targeted for the study, and 3) as a group that is open only to parents of children with PANS/PANDAS, its' members include families from multiple medical practices, families at various stages of their journey, and families with children across a range of ages. PANS/PANDAS Facebook groups typically include families and others who are affected by PANS/PANDAS and are used as a place to communicate about doctors, medication/treatment options, pose questions regarding a child's symptoms, and obtain support. The flier advertised the opportunity to participate and those interested were prompted to reach out via the email address and phone number provided. The flier can be found in Appendix A. After the initial flier post, some families and PANS/PANDAS advocates shared the flier widely across other networks, including group chats and other PANS/PANDAS Facebook groups.

Families of children with PANS/PANDAS can be considered a hidden and hard-to-reach population (Singer, 2013). Singer explains such populations as residing “outside of institutional and clinical settings,” (Singer, 2013, p. 255) as they often suffer consequences such as stigma and discrimination based on their condition. Locating these populations requires engaging with their networks (Singer, 2013). Further, in terms of sampling, these populations are relatively difficult to find and recruit which leads to a lack of knowledge of their characteristics and distribution (Singer, 2013, p. 261). According to Atkinson and Flint (2001), snowball sampling is particularly effective for accessing hidden and hard-to-reach populations. In this way, a sense of trust is built as referrals are made by peers or friends (including, in this case, other parents of

children with PANS/PANDAS). Social media, such as online platforms including Facebook groups and online parent forums, have given new opportunities for hidden populations to come together and create an atmosphere in which opinions and information are exchanged; also providing an avenue for researchers to recruit these hard to reach populations, as evidenced by the current study.

Procedures. Families interested in being a part of the study were asked to provide preliminary information, such as geographic location, child's current age, date of onset and date of diagnosis, and availability for an in-person interview to ensure they met inclusion criteria and to obtain a sample with adequate variability. Interviews with parents were then scheduled on a rolling basis. All interviews were conducted in-person with parents, for a total of 20 families with children diagnosed with PANS/PANDAS, living in the northeast of the United States. When possible, interviews were conducted with both parents present. Interviews took place at the home of the interviewee(s), in private meeting rooms at the families' local public libraries, or in private research and office spaces on campus. Interviews lasted anywhere from one hour and 52 minutes to four hours and 23 minutes with the average interview lasting two hours and 52 minutes. Each interview was recorded with digital audio recorders and transcribed verbatim with Descript, a transcription and editing program.

Ten undergraduate students at the University of Connecticut were responsible for reviewing the transcripts to ensure all identifying information was removed or edited and that the transcripts contained no errors. Transcripts were then reviewed and revised when necessary by the project manager to ensure agreement. In the case of discrepancies between the undergraduate researcher and project manager, the two conversed until an agreement was reached. Once transcriptions were complete, they were uploaded to the coding platform NVivo, for analysis.

Protection of Human Subjects. The current study was presented to and approved by the University of Connecticut Institutional Review Board (IRB) in Storrs, Connecticut. The IRB approval can be found in Appendix B. Prior to the start of each interview, parents were given a consent form detailing the nature of the study and measures taken to ensure confidentiality. This document can be found in Appendix C. Participants reserved the right to refrain from answering any questions or to withdraw from the study at any time. Additionally, participant families were compensated \$75 for participation in the study.

Participants. For the purpose of the current study, we reduced the sample from 20 families to eight by purposely excluding cases from analysis in which a child was given a previous autism spectrum disorder (ASD) diagnosis (even if misdiagnosed). This was done in order to analyze the impact of PANS/PANDAS in the absence of additional stressors that can accompany the navigation of the healthcare system and support services for ASD. Additionally, it was understood that families can have difficulty disentangling PANS/PANDAS symptoms from ASD related challenges and therefore, could obscure the experience of a PANS/PANDAS diagnosis on its own.

All eight families included in this analysis reported their racial/ethnic background as white/Caucasian. Parents were asked to report their education level, the distribution is as follows: 6.67% completed 12th grade or less; 6.67% received a high school diploma or GED; 26.67% had some college/Associate's degree/technical school training; 13.33% had received a Bachelor's degree; and 46.67% held a Master's or Doctorate degree (MD, PhD, JD). Six of the eight families (75%) reported their marital status as married, one reported themselves as single, and the other reported divorced status. Families were also asked to report their employment status: 80% of the parents in this study were employed full-time; 13.33% were part time; and 6.67%

were not employed. Family income was broken down into 7 categories, ranging from “Less than \$5,000,” to “More than \$150,000.” Three families reported their income as less than \$100,000 (\$5,000-\$19,999; \$50,000-\$69,999; and \$70,000-\$99,999 respectively). Two families reported their income as \$100,000-\$149,999 and the remaining four families reported their income as more than \$150,000. For the divorced family, incomes were reported separately. Three families reported using Medicaid/Medicare/ADAP/VA to pay for health care regarding their child; one of these families had Medicaid only, the other two had both Medicaid and private insurance. Seven families reported using private insurance and all eight families reported using self-pay or paying out of pocket.

Parents’ age ranged from 36 to 61 with an average age of 46. Children’s current age ranged from 7 to 22 with an average age of 12. The child’s age at onset ranged from 4 to 10 with an average age of 7. In this study, 40% of the children were male and 60% female. Five children had received a PANS or PANDAS diagnosis less than 3 months from their symptom onset; one child received a diagnosis within a year from onset; two children received a diagnosis 1-3 years after onset; and two children received a diagnosis 3 or more years after symptom onset. It should be noted that two families had two children with PANS/PANDAS, the remaining six families had only one child with PANS/PANDAS and additional children without PANS/PANDAS.

While some families were excluded from the final analyses, the eight families provided adequate data saturation. Methodological literature such as Hennink et al. (2017) and Guest et al. (2006), support the use of a smaller sample. Hennink et al. (2017) found that code saturation of data can be achieved within the first 9 interviews. Results of the Hennink et al. (2017) study showed that by the ninth interview, “the range of common thematic issues was identified, and the codebook had stabilized,” (p. 604). Further, Guest et al. (2006) found data saturation to occur

between seven and 12 interviews and many of the primary themes were present between interviews one and six. With this in mind, we feel confident in our sample of eight in-depth interviews based on the exclusion of those with an Autism diagnosis and reaching data saturation.

Phase Two: Data Analysis

An identical process to the one described for the parent forum threads was conducted for the analysis of the semi-structured in-depth interviews. In the initial phase, codes developed in the analyses of the parent forum threads (as previously described) were utilized when relevant. For these reasons, the analysis of the interviews had aspects of deductive thematic analysis but was largely inductive (Braun & Clarke 2006; Braun & Clarke, 2012). As the interviews were centered around the family's illness journey, information such as treatment trials were discussed but were not coded for the purposes of the current study. Instead, coding focused on the parts of the interview in which family dynamics, family functioning, and parent-child relationships were described. More descriptive codes were applied such as "parent always on alert" and "uncertainty in day to day functioning." These codes were developed through participants' language and concepts as well as from the researchers' theoretical interpretations (Braun & Clarke, 2012). In addition, the focused coding of the interviews served as an opportunity to check the accuracy of the initial codes and to confirm or deny any patterns seen from the online parent forum threads (Charmaz, 2006, p. 59). Surprisingly, the same codes were equally present across the online parent forum and interviews with the interviews largely operating as validation of the patterns seen in the forum. As in the analysis of the parent forum threads, themes from the interviews were identified. Comprehensive themes were then compiled after review and

organization of the themes that best represented the diversity of and patterns within both the online parent forum and interviews.

RESULTS

It should be noted that there was great overlap and congruence in the experiences described by both parents from the forum and parents in the interviews. Accordingly, results reported below reflect the integration of the interview and parent forum analyses.

Through the analyses of the online parent forum and eight in-depth interviews, we have come to understand the distinct ways in which family functioning is compromised when a child has PANS/PANDAS. The interviews and forum differed in the types of narratives provided, with families describing their entire journey retrospectively in one long meeting whereas the forum is longitudinal with parents posting their experiences as they occur in real time (although some posts also reflect back on the journey). However, patterns identified in each were validated by results in the other. With this in mind, it has been found that families on the forum (which are anonymous) self-disclosed in a similar way to families participating in the in-depth interviews. Given the congruence and lack of any conflicting results between the two data sources, four primary themes derived from analyses of both data sources were recognized as contributing to the impact of PANS/PANDAS on family functioning for families with a diagnosed child. Some themes had greater saturation than others; the results presented here showcase the patterns that existed across many families. The four themes are reported below and include: a) Determinants of Family Functioning (sub-themes: child symptoms and clinical course & family resources and institutional barriers), b) Close Relationships (sub-themes: family sub-systems and support

network – extended family and friends), c) Routines and Daily Life, and d) Family Emotional Well-Being. A concept map can be found in Appendix D.

Determinants of Family Functioning

Child Symptoms and Clinical Course. As previously described, one of the hallmark features of PANS/PANDAS is its' relapsing-remitting nature of symptoms. This jagged pattern allows for times of complete dysfunction as well as relative calm for families. It was found that depending on the state of the child (i.e., well or in flare/exacerbation) the family's level of functioning fluctuates as well. When the child is well or currently not suffering from a heightened level of symptoms, families describe themselves as living a largely normal life. Routines that existed prior to the onset of PANS/PANDAS are resumed in some form and the child may return to school after continued absences or begin involvement in an extracurricular activity that had been suspended. Similarly, parents may return to a job that they had taken leave from. Parents also described these periods of time as their child being "back," indicating that when the child is in flare, they are not their typical selves sometimes to the degree that they describe their child as "lost," or "gone." It was found that parents employed the use of percentages to describe their child's level of functionality or return to a pre-PANS/PANDAS state ("up until now, she's been 100%", "back down to 50% functional", "within three or four days, she was 90% improved"). Parents utilized this strategy of quantifying their child's functionality as a means of tracking whether treatments were working and/or deciphering if the child was in the remittance phase. Months or even years can pass between each phase, leaving families with the strain of not knowing if another exacerbation is upcoming. Families expressed the challenge of waiting to get an appointment with doctors or specialists while their child is

experiencing an exacerbation. One parent describes the relapsing-remitting pattern and uncertainty that comes with it as follows:

Jennifer, interview: Then she started doing well...that's the thing. It ebbs and flows. So you're like, "Okay, she's kind of good." Or, "Do we really go to this appointment now?" Sometimes when you go, they're in a good place, 'cause you're waiting for this appointment, and you look insane. Because they're probably like, "Alright these parents need to get a life." ... [their kid] Is perfectly fine.

In periods of relative calm, families described their ability to manage the child's needs and treatments and tended to have a more positive outlook.

There is also a great level of variability in the severity across children with PANS/PANDAS and in their recovery level. Parents of children with PANS/PANDAS have the challenge of deciding how to handle behaviors that were non-existent in their child at one point in time but were now taking over family life. Some children are considered to have mild symptom severity, often "flying under the radar" because their OCD rituals did not involve externalized behaviors but intrusive thoughts. In addition, parents were not always perceiving a child's behavior as a signal of an underlying problem but rather as something "quirky," such as constant eye blinking, being very organized, or existential worries. In sharp contrast, some children with PANS/PANDAS engage in often involuntary, yet frightening rages where both the child's and families' safety could be in jeopardy. These can involve the child trying to hurt others or themselves with some parents describing that their child has put holes in the walls or broken other items around the home during a rage episode. In four of the eight interviews, parents detailed an incident in which the child grabbed a knife or remarked they wanted to hurt someone or themselves with a knife. With this in mind, each child's trajectory for recovery is variable as well. Several children are able to get back to "100%" in the sense that they are no longer impacted by PANS/PANDAS symptoms. Others have residual mild symptoms, that with

continuous management, they are able to move forward in life. A portion of children though, remain severely affected over time. In these cases, some families must settle for symptom management rather than a return to their pre-PANS/PANDAS state. The variability in severity and recovery, as well as the relapse-remittance patterns, requires families to continually adapt their strategies for maintaining family functioning and well-being.

It was found that for a large number of families affected, their journey spans across multiple years. It was not uncommon for families to experience a multi-year lag between the onset of symptoms and an official diagnosis. This time period may be the most difficult and the one in which most of the disruption to family functioning happens. After diagnosis, families continue to combat the ups and downs as the child approaches puberty and occasionally into adulthood. For these reasons, parents indicated that those with PANS/PANDAS are robbed of their childhood. Given the relapsing-remitting nature, parents described the fear of relapse and need to be vigilant to quickly treat any symptoms that may appear. The challenges that alter family functioning are therefore long-term often without a clear point at which the child can be deemed “cured.”

Family Resources and Institutional Barriers. The variability in the child’s symptoms and clinical course described above can also lead to changes in access to resources for the family. One of the major strains on the family’s resources was lack of support from the medical community. Parents frequently described the first obstacle in discovering the cause behind their child’s dramatic change was finding a doctor who was able to accurately diagnose their child’s illness. Children with PANS/PANDAS are often diagnosed with Tourette’s syndrome, childhood OCD, Pervasive Developmental Disorder and others, before achieving the correct diagnosis. In this process, parents were often turned away by doctors and medical professionals who have not

heard of PANS/PANDAS or were aware of it but did not believe in it. Parents were found to be highly stressed and concerned for their child's well-being as the child would continue to suffer from symptoms. Parents from the forum and interviews also remarked that it was a doctor's belief in the parent and in PANS/PANDAS as the most helpful piece in their journey. After being turned down repeatedly, parents felt that even if a doctor was unsure how to treat their child, simply listening to the parents' concerns was monumental.

Further impacting access to resources was the experience of a parent having a change in their career in order to become a full-time caregiver for their child with PANS/PANDAS. The impact of this change manifests in a decreased income for the home which in turn affects the ability to afford doctor's visits, medications and procedures, and other expenses involved in taking care of the diagnosed child. It was common for parents to remark on their financial situation and relevant expenses when describing their journey. Often, parents explained their emotional struggle as largely influenced by the great financial burden. For some families, there was reference to extreme financial losses including losing a home and filing for bankruptcy. Others were able to afford the necessary medical attention without going into debt. The majority of families were in the middle, experiencing a blow to their finances but were fortunate enough to remain financially stable.

Acquiring a PANS/PANDAS diagnosis requires specialists or informed pediatricians willing and capable of treating, both of which are still sparse in the medical community today. Appointments and treatments though, are rarely covered by health insurance plans. Additionally, a child is hardly ever treated by a single doctor but instead sees multiple to handle the various aspects of the chronic illness and variable responsiveness to treatments. For example, the family may see a PANS/PANDAS specialist to confirm diagnosis and start treatment, but will work

with another in regards to treatment of other infections or to augment with alternative treatments. Still, other families have the child see a therapist as part of their medical “team.” Interestingly, some families acknowledged that while they were able to pay for the doctors and treatments, the financial burden was large and were aware of the hardship others families might have if not financially able. The level to which a family’s finances were affected, dramatically impacted the child’s access to medical attention and could change the chances of returning to full health. The examples below speak to the reality that many families face.

Tracey (mother), interview: We just got his therapist right now...I've called all the doctors...And she said [receptionist at therapist office] they have a wait list, but she was, in the meantime, she was going to talk to the doctors about things and see if there was anything they can do to help us.

Paul (father): Because right now we're at a standstill.

Tracey: Nobody takes insurance...We're at financial end at this point.

Andrea, interview: But I ended up filing for bankruptcy because I was charging, you know, a lot of stuff to it [credit card]. I spend thousands on supplements.

Parent 1, forum: The money we have spent trying to diagnose, treat, and get services for help with this disease destroyed us emotionally, physically, and financially.

A similar level of frustration was found when families lacked support from the child’s school system. Refusal to work with parents on accommodations, disbelief in the diagnosis, and removal of the child due to high absences, left families struggling to maintain their child’s academic trajectory. It was found that families with a child diagnosed with PANS/PANDAS would often ask the school to inform them if another student was sick so as to prepare for a flare, but this request wasn’t always upheld by the school. As children with PANS/PANDAS tend to experience a decline in both behavior and academics, including handwriting and mathematics comprehension, they may come to be regarded as problematic in the classroom. Additionally, parents experienced teachers making remarks that their child’s behavior was solely due to “bad parenting” or “lack of discipline.”

While parents reported many struggles with their school system, positive experiences included, the provision of a tutor or materials for when the child is absent due to an exacerbation, effective execution of and commitment to accommodations as outlined in an Individualized Education Program (IEP) or enactment of Section 504, and overall acceptance and compassion by teachers and faculty. A few families remarked that in time, the child no longer needed certain accommodations but kept them in place as a safety net. In some exceptional cases families praised the school system, for without their assistance, their child would not have been able to maintain their academic trajectory.

The determinants of family functioning (child symptomology & clinical course and family resources & institutional barriers) are considered to be determinants as they represent the basis from which a family's experience proceeds. For example, the experience of a child's severe symptomology, multiple years between onset and diagnosis, limited financial resources, and difficulty obtaining treatment likely contribute to greater family distress than for families whose child has milder symptom presentation, quick diagnosis, higher financial stability, and successful treatment progression. The variability in each of these determinants showcases that while families of children with PANS/PANDAS are not alone in their journey to return their child to full health, the path is not universal.

Close Relationships

Family Sub-Systems. A family is made up of its component parts, including the parent relationship (marriage and co-parenting), the parent-child relationship, and the sibling relationship. Analysis of the forum and interviews revealed trends in these relationships that ultimately contribute to the family's ability to maintain a level of functioning with a child with PANS/PANDAS.

Parent Relationship (Marriage and Co-parenting). It was found that the parent relationship was greatly impacted by the introduction of this chronic illness in the child. Parents discuss their marriage being tested in times of stress with some ending in divorce. Though it could not be confirmed by the username on the forum, comments indicated that the user was more often the mother than the father of the child. In interviews in which both parents were present, it was again the mothers who had taken on the role of researcher, appointment maker, and overall caretaker for the child. Fathers, consequently, tended to uphold the role of provider by maintaining their job or increasing their hours in order to bring in financial assistance needed to pay for medical bills or treatments.

Another common dynamic between parents was the father remaining skeptical or taking more time to come on board. There appeared to be a split between supportive and engaged fathers, and those who were distrusting in the diagnosis, or removed themselves from the family dynamic. One of the factors that contributed to the strain or ultimate dissolution of the marriage was the pattern of parents taking on different roles. It was uncommon for both parents to equally spend time researching as well as participating in doctor appointments. For some families this division of tasks was a conscious decision whereas others fell into the roles without much thought or consideration. The example below illustrates this issue of miscommunication.

Parent 2, forum: I have always felt knowledge is power - so once I had a name for what was wrong with my kids - I set out to learn everything I could and educate everyone dealing with our kids. My husband on the other hand, felt he had little control over the medical side and it overwhelmed him - so he tried to obtain control in his way - financially. He set to rearranging our finances, investments, etc., so he could pay for whatever doctors, medicine, treatments etc., I said were needed without bankrupting us. It was his way of maintaining control. That he did this without a word to me was part of the problem. What I saw as him being aloof and uncaring was really just him dividing and conquering.

Across the forum and interviews there were stories of divorce, often leaving one parent to take on both the provider and caretaker roles. In this situation, parents found it increasingly more difficult as they are not afforded a “break,” from their child and were forced to make decisions without council from a spouse. In instances in which a divorce occurred, the two parents then dealt with the issue of co-parenting. It was seen that again, one parent tended to take on both the provider and caretaker role while the other parent was uninvolved. Still, for other families, parents remained married but had separated emotionally (“Our marriage is essentially over but we are still partners in raising our children”).

Another factor that was found to impact the parent relationship were instances of disconnect between the parents regarding treatment, help needed, the existence of PANS/PANDAS, and the ability to move on after the child has achieved full health. Many instances across the forum and interviews pointed to the pattern of parent disagreement on the treatment protocol for the child. Some treatments such as IVIG and plasmapheresis do not guarantee that the child will return to full health and can be very costly as they are typically not covered by insurance. For these reasons, parents may debate if the treatments are worth the financial and clinical risks without knowing if the treatment will work. In some cases, this was further complicated by differences in the child’s perceived severity and belief in PANS/PANDAS as a whole. The following excerpts help to illustrate these contrasts.

Jennifer (mother), interview: But I also in the beginning felt like I was always trying to convince him, this is what it is. Because you weren't really the researcher, at all.

Steve (father): No.

Jennifer: So I was doing all this research, and then I felt like as time went on, he would have these big opinions about it, but he wouldn't have done any of the work to formulate that opinion. So it would make me mad, and then we would get into an argument. And then if I wanted to try this route, like the naturopaths, he was like, "Herbs?" like, "What's this?"

Steve: That's the way I confirm it is I'll challenge it by researching that, but on the opposite side, to get opposite opinions.

Vanessa, interview: And I had in the meantime broken the court order. And I got her an appointment with Dr. T. Which I wasn't supposed to do without his [ex-husband's] permission but I said, "You know he's not going to give me permission for anything." And I said, "You know what, what the hell, he can hold me in contempt but my kid has to come first." And so I had called him and I said, "Look, file a contempt motion if you want to file it but I made an appointment for Emma." ... "That's the first person that can see her. This is this PANDAS thing, I sent you information." I already sat down with him and his wife about it and they're telling me that, "No it's not real. It's a figment of your imagination, this doesn't exist you just want us to pay money."

For the families who described that their child had been symptom free for an extended period of time and believed that the child was no longer affected by PANS/PANDAS, parents faced the task of living a “normal” life. It was found that parents engaged in different ways of coping with their new found freedom after years of dysfunction. Fathers tended to explain a readiness to move on and leave the trauma behind. In contrast, mothers followed a pattern of continuing to feel the effects of the intense caregiving. Given the multiple areas of disagreement that can happen in the parent relationship, families pointed out that their marriage was greatly changed and lacked connection due to years of neglect in favor of the child’s well-being. To help navigate the new territory, families discussed the use of marriage counseling to process the experience and regain connection to each other. For families in which the father was quick to acknowledge and accept the child’s PANS/PANDAS (versus skepticism), this fact appeared to be an integral piece in the survival of the marriage. Two parents describe how attention was diverted from the marriage and needed to be put back after the child is well:

Tracey, interview: And just, we don't get any time. Our marriage is-- I feel like it's kind of in trouble. We don't get any time together. We don't get to go out together. We can't speak to each other without him being right there, butting up into everything.

Parent 3, forum: I also think that my husband and I need to then go back and work on

the collateral damage to our marriage. There is no way a child can feel whole when a husband and wife can hardly recognize each other anymore.

Parent-Child Relationship. Through the analyses, patterns of alterations to the parent-child relationship were also found. It was found that parents experienced periods of devastation and confusion. For example, along with the experience of depression, panic, and anxiety, parents made comparisons to the life they used to have, as well as to how their child used to be (“straight A student to IEP special Ed.”). Parents referred to “grieving the ‘loss’ of their child,” as often years have passed since they have seen their child developing on a typical trajectory. Even after the family was aware that their child had PANS/PANDAS, the parent-child relationship continued to shift. In many instances, parents spoke of their child’s severe separation anxiety, making it difficult for the parent to complete tasks or leave the house. For some parents, this spurred feelings of resentment and wanting space from their own child. Parents had difficulty admitting that they fantasized about a different life or returning to the one that they used to live. In addition, a common opinion was the thought of “getting in the car and driving away.” Likewise, parents experienced mixed emotions in regards to the child’s obsessive-compulsive behavior or tics. A trend was noted that several parents could recall one point in time when they felt annoyed or embarrassed by these behaviors. These parents would then acknowledge their fear of being labeled as a “bad parent,” for having such emotions. In addition, analyses revealed that typical parenting practices may not apply when a child has PANS/PANDAS and that parents of children with ongoing symptoms often report revising expectations and long-term goals for their child’s future, such as college or becoming independent and living on their own.

Further complicating the parent-child relationship were instances in which the child was removed from their regular schooling and transitioned to homeschooling. In multiple cases

across the forum and interviews, it was the primary caregiver who then became the homeschool teacher. In these cases, parents described having even less time to themselves and difficulty taking on another role as teacher. For other families, it was not the homeschooling that caused a strain in the parent-child relationship but the rages that their child experienced. It was found that parents and other family members, such as siblings, were the target of aggression by the child with PANS/PANDAS. Parents experienced fear in trying to protect their other children and themselves when a rage occurred in the home. In these instances, parents had to consciously remember that the child could not control their behavior and the child was often not fully aware of their behavior; some would even fail to remember these episodes later. Families on the forum and in the interviews detailed the challenge of trying to stifle their natural fight or flight response when the child was in a rage.

In sharp contrast, some parents regarded their experience with PANS/PANDAS as an opportunity to grow closer to the child in a way they would not have otherwise. Across both the forum and interviews, some parents were able to positively reflect, indicating that they learned important skills such as distress management, patience, and empathy. The ability to assign a positive lens to the experience was found to occur more often when the child had reached a steady state of health and after the family was able to regain some functioning in their daily life.

Sibling Relationship. In the forum and interviews, there was an overwhelming concern with the effects of having a child with PANS/PANDAS on the other, healthy, children in the family. Of greatest concern was the mixed messages healthy children received when their PANS/PANDAS sibling did not suffer the same consequences for unacceptable behaviors. Parents were open to admitting that they often asked their healthy children to make accommodations for the child with PANS/PANDAS in ways such as allowing time for an intense

eruption of symptoms (OCD rituals, tics, rages) to subside or missing out on activities because it would cause a problem for the child affected by PANS/PANDAS. In addition, parents were not oblivious to the fact that their attention was unequal across their children. As one parent describes:

Parent 4, forum: It's so tempting to ask the healthy one to "suck it up" and compromise while you deal with the crisis child.

Parents were also found to express guilt and concern about the violent and/or traumatic episodes that the sibling witnessed, especially if the sibling was young. The siblings are sometimes thought to live an alternate life as the parents try to remove them from difficult or harmful situations involving their PANS/PANDAS child and to maintain a level of normalcy. Some families believe that their healthy children learn to become independent and apply a positive lens, reflecting on lessons they have learned about being tolerant of those with illnesses or disabilities. However, many recognize the neglect that their healthy children experience across time:

Parent 5, forum: I do think the "typical" siblings suffer more than parent[s] know and, though it's hard to do during the chaos, [they] need help too.

For these reasons, parents tended to view the relationship between the children in the home as altered or strained.

Support Network – Extended Family and Friends. On both the forum and in the interviews, families described the tendency to not discuss their child's diagnosis with many others in their lives. Additionally, some noted that they actually lost connections with family and friends due to their child's illness. Parents found it difficult to maintain relationships, particularly when others did not understand their challenges, and felt the need to avoid situations they knew would elicit reactive responses from their child. This included consistently turning down

invitations to birthday parties and gatherings for fear that their child would become symptomatic. Families also expressed their frustration when friends or family would blame the parent for the child's behavior, attributing it to an "issue of discipline."

However, it was also found that the online parent forums and Facebook groups, specifically for families of children with PANS/PANDAS helped to fill the gap, providing families with a dependable support network. In the forum, parents will sometimes share very vulnerable information, such as the need for space from their child, as previously described. Likely due to anonymity and the knowledge that other parents have similar experiences, parents discuss freely without fear of judgement. Also, as evidenced by the nature of the thread posts, support and encouragement are offered even in times of severe distress. Occasionally, parents continue their conversations offline and work to organize support groups in person in their area. While some families may experience the loss of close friends and family due to their unique situation with PANS/PANDAS, they are often able to gain support from these online communities. Below are two examples that demonstrate the difference in support families can experience.

Vanessa, interview: So I found that a lot of parents say the same thing, you ended up with a much smaller circle, but the people that are in your circle are really solid. And that's a blessing.

Tracey, interview: And they [friends] get so tired of hearing about all your depressing crap, they stop asking how you're doing, and then it just all stops.

Though close relationships such as the ones described above may appear to be peripheral in their influence, the current results suggest that such relationships can act as either direct hindrances or buffers to family functioning.

Routines and Daily Life

One of the more difficult challenges to family functioning is alterations to previous

routines and daily tasks. In particular, when the child engages in school refusal or must be homeschooled to avoid consistent exposure to strep and other illnesses that will trigger an exacerbation of their symptoms. As previously mentioned, the family must reconfigure, typically resulting in one parent being forced to stay home with the child. It was also found that some children are not enrolled in homeschool programs and instead miss large chunks such as “30, 40, 50 days,” or whole years of schooling when symptoms are severe. The decision to switch to homeschooling or simply remove the child from the school is often a result of the aforementioned separation anxiety that many children with PANS/PANDAS experience. It can be an everyday battle for the child to get to school. The child will often engage in school refusal due to fears associated with the school such as the fear of vomiting in the classroom or sensory challenges such as not being able to wear clothes because they are uncomfortable on the child’s skin. It was found that in the event that the child can attend school in some capacity, accommodations are made including an IEP or 504 plan. Some parents reported having arguments with the school to acquire the necessary means to keep their child following their academic trajectory as best as possible. Depending on the school district, its understanding and compassion towards the child’s diagnosis, and willingness to help the families make accommodations, were large factors that impacted the family’s ability to keep a child in school or not.

Many families also described the need to make changes in the child’s diet at the urging of practitioners and other families on the online parent forum to increase the child’s chances at returning to full health. It was found that families reported some success and improvement in their child’s functioning after switching to a gluten free, soy free, dairy free, or related diets. To show support for their child, families tended to make these adjustments for all members in the

home and not just the child with PANS/PANDAS. Families are encouraged to abide by such dietary restrictions in conjunction with strict treatment regimens in the form of antibiotics or herbal remedies. However, these protocols were found to be quite complex, expensive, and time consuming. For instance, parents described the need to sometimes give their child three different supplements at three different times throughout the day. It is thus understood why some families then choose to keep the child at home to best monitor symptoms and administer treatments as necessary.

The demands that come with the strict treatment regimens paired with the inability or refusal to attend school was often found to require a parent to make adjustments to their career in the form of part-time or reduced hours and in some cases, quit entirely. This loss of a job to allow for the maintenance of their child's health was particularly difficult for families, across both data sources as shown in the following examples:

Interviewer: You mentioned you work.

Tracey: I'm not anymore. I had to quit my job...because I was going to have to [provide] transportation to the [partial hospitalization program]...So that would leave me no time to go to work.

Jessica (mother), interview: I also went part time that was another thing that we had.

Mark (father): She actually cut down from being full time to part time just to help, manage the situation so, um, to have a bit more time.

Parent 6, forum: I cannot work- had to quit a job that I loved- because with two [kids with PANS/PANDAS], it seems that one is always either not in school or on the edge.

It was seen that the primary caregiver begins a new "full time job" as they serve as more than the typical parental provision of love, shelter, and safety. This position was explained as having to schedule and attend multiple doctor appointments both near and away, maintain the child's treatment regimen and/or diet, manage the child's behaviors and psychiatric symptoms, conduct online research, and possibly teach their child's homeschooling program. However, leaving a job

was typically not feasible when the primary caregiver was a single parent. For the families in which a member had lost their job or had to leave voluntarily, the parent tended to lose a part of their identity. Many remarked that they loved their previous jobs and it was not always an easy choice to make the switch to stay-at-home parent and full-time caregiver.

Another element that was found to make an impact on the daily life of these families was the experience of feeling isolated from friends and family as they would often have difficulty leaving the house. This inability to continue daily functions involving places outside the home was often due to a lack of family or babysitters who could handle the child's behaviors and needs, a child's separation anxiety, the child's heightened reactive responses in public spaces and/or fear that the child will get sick and begin a flare. The following examples illustrate the level to which the family sacrifices their daily functioning to accommodate for the child.

Parent 7, forum: It is so tough, and with these really treatment-resistant cases, the feelings of helplessness and isolation are nearly as devastating and debilitating as the disease itself.

Parent 8, forum: We are unable to go out unless we have one on one coverage for him. I leave him at home with our fantastic sitter. It is hard because I want him to learn how to behave, but when he is sick it is hopeless.

Tracey, interview: So, we don't, you know, we don't get to go out and go to the store or anything like that...the only other time we've been able to get out was a funeral.

It was found that parents described how their world came to revolve around their diagnosed child or how the family life had "gotten out of control." It should be noted that there were certainly instances in which such accommodations were not necessary, for example, both parents being able to remain at work. However, each of the alterations to daily life discussed above (homeschooling/school refusal, diet and treatment regimen, loss of job, and isolation from family and friends) indicate the often overwhelming and consuming nature of having a child with PANS/PANDAS.

Family Emotional Well-Being

The abrupt onset and severity of symptoms of PANS/PANDAS indicates a level of hardship that is distinct from families with children who develop as anticipated or for families of children with chronic, but widely accepted and manageable conditions. Families of children with PANS/PANDAS are distinctly stressed with extreme emotional and behavioral symptomatology that is not characteristic of most childhood chronic illnesses. One of the most devastating impacts is the parents' feeling of uncertainty that accompanies a family's journey with PANS/PANDAS. This uncertainty manifests in the child's variable day to day functioning; the unknowable future of the child's health, development and well-being; and the lack of a clear course of action in regards to treatment and the process of returning the child to full health.

Below are some examples from the online parent forum and interviews describing the multiple facets of uncertainty.

Parent 9, forum: I so was hoping for the doctors to tell me what path to take, and what would heal our son...but to no avail. There are so many triggers, genetic predispositions...and so many different routes for recovery.

Parent 10, forum: We worry everyday...and with each strep encounter and flare, however small it may be, we wonder IF and WHEN we will be past this.

Interviewer: Thinking of everything we've talked about. What is PANS for you?

Mark (father): Um, not knowing. Not knowing what's coming our way...not knowing, what the day's going to hold, behavior wise. Um. And. Um. I guess dreading.

Vanessa, interview: You know if she wants to have children someday. Who knows how pregnancy is going to be. You know these are all the things they don't know about PANDAS kids who grow up...it's like going in this very unknown territory...How do they do in pregnancy? Are there any risks? Is there anything they should do?

Jennifer, interview: You feel scared too 'cause you don't feel like there's a doctor, someone that you can go to. You just-- you feel kind of alone in it...I wish it was a disorder that's like, like a broken leg. You know how to fix the broken leg. And it's not like that. And when you feel like some doctors don't believe it, and you just feel alone a lot. Like, "Who do I call?"

Magnified by the uncertainty that surrounds the experience of a child with PANS/PANDAS, parents across the forum and interviews express feelings of caregiver burden. This included: (1) the sense of always being on alert for the presence of symptom return, (2) parent burnout, (3) deteriorating mental and physical health of the caregivers, and (4) various emotional responses such as devastation, frustration, and feelings known to be indicative of post-traumatic stress disorder (PTSD). Families portrayed their daily life as a state of hyper-awareness so as to be prepared to act should their child begin a phase of exacerbation. A few examples of this state of hyper-awareness are below.

Parent 11, forum: I get so tired of being constantly on alert, trying to make sense of what I'm seeing with DS [dear son], trying to stay one step ahead, having discussions with our new family doctor about the right antibiotic...constantly reading and researching, talking with each of the members of our team, micromanaging...but my head goes right back to a year ago when we were in crisis. Talk about PTSD!

Mark, interview: For us it's like, we're still wired ready, ready, we're like we're ready for the something to happen so it's like for us, we're never resting.

Jennifer, interview: It's always on your mind. You're always basically looking for symptoms. Is that it? Is that not it? And you're looking at other children—your others kids too.

Parents were found to experience feelings of burnout and exhaustion as they often have to physically assist their child in daily tasks. As children with PANS/PANDAS can lose skills that were previously acquired such as handwriting abilities, math competency, and motor dexterity, parents must step in to assist with these tasks. The parents in the interviews and the forum frequently spoke about the struggle of getting their child bathed, dressed, and out the door to school or an activity on time due to the child's OCD or tic behaviors. Additionally, parents were quick to point out that tasks such as homework, took much longer for their child than those at the same grade level without PANS/PANDAS and would often require a parent to keep the child on task until complete. Similarly, a child's OCD symptoms might include participation by the

parent such as the parent having to repeat a phrase to the child or provide reassurance repeatedly. Parents also described the feeling of taking on their child's anxiety or experiencing panic attacks, for example:

Tracey, interview: And of course me and him [child with PANS/PANDAS] feed off each other and we bump heads...and now his anxiety gets my anxiety up.

Across both the interviews and forum posts, it was common for the mother specifically, to be an advocate for their child; researching new treatments and options for their child to thrive as they would without this illness. In taking on this new role as caregiver of a child with a chronic illness, parents expressed sometimes seeking medical treatment for themselves in the form of anti-anxiety medication or talk-therapy with a psychologist to process the heavy emotions and physical exhaustion.

The demands that this chronic illness places on the family takes a toll, with parents specifically remarking on their lack of sleep across months or years. Unfortunately, lack of sleep is not the only sacrifice, as parents also described neglecting their own health for that of their child culminating in weight gain, increase in symptoms of a previous health condition, or introduction of a new health condition (such as breast cancer or high blood pressure). As the primary caregivers for a child with PANS/PANDAS, parents described the experience of being the child's "safe person." Due to the fact that the child saw the home and parents as a safe space, their symptoms would tend to manifest more at home. There was often a disconnect between reports on the child's behavior from the school teacher and what was experienced in the home. Parents believed that the child would "hold it together" during the school day only to fall apart upon returning home. On the other hand, some children were not able to suppress their behaviors in school. Parents commented on the times they received phone calls from the school that their child had been aggressive or could not sit still for an activity. Even in those cases, the worst

symptoms were still experienced at home by the parents and siblings. For these reasons, parents carry the burden of managing their child's symptoms that others might not see.

Throughout the families' journey, parents experienced deep and intense emotions similar to PTSD, with a few parents acquiring an official diagnosis of PTSD. Even in families who had seen sustained improvements in their child for long periods of time, parents still felt strong emotions in the aftermath. The uncertainty, as previously described, and the emotional drain experienced during the ups and downs contributes to the pervasive nature of PANS/PANDAS and what many families describe as a traumatic experience.

Analyses revealed frequent use of language indicative of trauma (e.g., "PTSD," "this is war," "torture") to illustrate the experience of having a child with PANS/PANDAS. As an example, one parent from the online forum wrote:

Parent 12, forum: Your PANDAS experience seems like a war in which you're enlisted without having known it. Somebody drops a bomb on your house one night, and there's no turning back. And when that bomb hits, and for the period after the primary destruction, it's a horrible mess. There's no rest, no solace, no normalcy.

Similar metaphors equating the experience to war were common across both the forum and the interviews, such as "going to battle" and "fighting to get my child back." It was found that parents also employed the use of otherworldly metaphors to illustrate their child's behavior including, "living with a terrorist," "like a zombie" (when on certain medications), and "exorcist-like behaviors." Patterns emerged across interviews and the forum equating the entire experience as a "nightmare," or "hell." The following examples illustrate this pattern:

Parent 13, forum: Until this disorder hit, we were a very peaceful, respectful, and loving family. Now there are days I scream at my dd [dear daughter] and say mean things back to her when she just can't help herself. It's like I've fallen into a nightmare and cannot wake up.

Parent 14, forum: This post throws me back into the "nightmare days" when our house felt like a war zone and my wife and I were totally shell-shocked. Our son (like many on

here) had been such a mellow, pacifistic kid before PANDAS began tormenting him, then that all changed during his exacerbation. Can hardly bear to dredge up those memories.

Interviewer: So, after all this discussion that we've had, what is PANS for you?

Tracey: Hell. Living hell.

Further, when families described the more difficult times in their journey, it was often referred to as a "crisis."

Such metaphors indicate that the experience is a dramatic change in the family's ability to function and requires accommodations in multiple areas of the family life. Interestingly, all of the metaphors explained above were not unique to one particular forum user or interview. The same terminology was found across both, indicating uniformity in the experience and the formation of a language only other PANS/PANDAS families could truly understand.

DISCUSSION

Families of children with PANS/PANDAS are decidedly affected by the extreme emotional and behavioral symptomology that is not characteristic of other childhood chronic illnesses. For these reasons, the current study sought to begin an important conversation and illuminate the experience of this particular population. Previous literature suggests impacts to family functioning, but does not address the specific ways in which family functioning is affected when a child is diagnosed with PANS/PANDAS. Through the use of both the online parent forum posts and in-depth interviews, the current study was able to fill the void in the literature on family functioning and PANS/PANDAS.

Findings from the current study did follow patterns of previous literature regarding children diagnosed with a chronic illness such as asthma, cancer, or type 1 diabetes, but with some distinct differences. For families affected by a child diagnosed with a chronic illness, including PANS/PANDAS, it was reported by Tona et al. (2017) that the child's functioning can

become impaired when a child is in exacerbation. Though not the aim of the current study, similar results were found. In fact, some parents in the online parent forum and interview phase saw such change in the child's abilities (for example, deterioration in handwriting) as an indicator of relapse and the need to begin treatment again.

McClellan and Cohen (2007) explained how the impact of the child's illness would vary in severity, culminating in mixed results as not all families with chronically ill children reported compromised functioning. This variability in family functioning was also a major theme of the current study. However, results of the current study had a clearer tendency toward reports of compromised functioning at multiple points in time. Only a few families from the interview phase were able to reflect on their journey and recognize times in which functioning was maintained; others were still in the crux of their journey and could not recall periods of time in which family functioning was not compromised. In addition, it was seen that after the child had achieved greater health, some parents stopped posting and responding on the forum as they regained functioning and began the next phase of life.

As families adjust to their new routines post-diagnosis, involving strict treatment regimens and limited time to participate in other activities or work, families tend to experience a loss of freedom (Coffey, 2006; Gannoni & Shute, 2010; McClellan & Cohen, 2007). High levels of stress and constant worry were also common emotions found in families of both PANS/PANDAS children and other chronic illnesses (Coffey, 2006; McClellan & Cohen, 2007). Notably, the stress and worry of families affected by PANS/PANDAS was often the result of the chronic illness' impact on all areas of the child's functioning and the distress of watching their child struggle. Gannoni and Shute (2010) discussed the impact to schooling specifically in that children affected by a chronic illness experienced more frequent school absences. This trend was

also found in the current study as families navigated the options of homeschooling (often requiring a parent to stay home and teach), partial day participation, and accommodations through IEPs or 504 plans. The constant worry as mentioned in Coffey (2006) was found to be more specific for parents of children with PANS/PANDAS due to the illness' relapsing-remitting nature.

The effect on siblings was another variable that often contributed to the heightened worry and stress levels. Coffey (2006) and Gannoni and Shute (2010) both described the fact that siblings often experienced a disruption to their social activities. In addition, at times when parents were able to reflect on their experiences, the possible trauma inflicted on a sibling was of primary concern. For families with a child diagnosed with PANS/PANDAS, siblings can be a target of aggression and rage which is noticeably distinct from witnessing a sibling navigate primarily physical symptoms of illness. In the critical review by McClellan and Cohen (2007), it was noted that the studies reviewed did not include siblings in the analysis of impact of child's illness on family functioning though they undoubtedly play a role that requires further investigation.

As Popp et al. (2014) found, parents of children with PANS/PANDAS are also split between those who are resolved or unresolved. Though not specifically addressed in the current study, nuances of a families' classification as resolved or unresolved were present in both data sources. It is thought that the inability for a PANS/PANDAS parent to reach a resolved state could be due to the stark, overnight shift in their child's demeanor. The age of onset varies for children with PANS/PANDAS and it is currently still unable to be detected from birth. Families can thus create bonds with their child that are shifted and changed if onset is later on in childhood. McClellan and Cohen (2007) posited that families who had ample time to come to

terms with a child's diagnosis with a chronic illness may have returned to functionality. This cannot be said with certainty for families of children with PANS/PANDAS as the ability to return to functionality was found to be largely affected by the family's resources, institutional barriers, and relapsing-remitting nature of symptoms.

Previous literature has spoken to the experience of caregiver burden as a common variable for families with a child diagnosed with a chronic illness. Frankovich et al. (2015, 2018) and Farmer et al. (2018) utilized the CBI to quantify the level of caregiver burden and the point at which parents exceed the threshold used to determine respite need. In the current study, caregiver burden was understood through in-depth descriptions of family functioning, impact on family resources, and impacts to routines and daily life. Though families of children with PANS/PANDAS consistently discussed feelings of caregiver burden, outside care was not found to be incorporated. Parents instead, focused on the need for help from the medical community and school systems, rather than an in-home nurse or caretaker. Caregiver burden was considered across five categories in Frankovich et al. (2018) and Farmer et al. (2018): time dependency, emotional health, physical health, social relationships, and development. The current study found indeed, each of these areas are factors to the experience of caregiver burden for PANS/PANDAS parents. Most notably was the decline in parents' emotional and physical health. As previously discussed, parents lacked sleep, were not typically involved in exercise, and were consistently in fight or flight mode due to the need to provide 24-hour care and assistance to their child. Similarly, parents of children with PANS/PANDAS felt isolated from previous social activities and members of their community. Some families described this as partly due to the fact that extended family and friends "don't get it." Development was not universally attainable for the families studied, especially in the interview phase. Despite the child's achievements with proper

treatment, some parents' descriptions of their journey would conjure up emotions as if the child had been recently diagnosed.

It is more often the mother of the child (rather than the father) who takes on the challenges that affect their child's daily functioning, as described by Coffey (2006). Feelings of being overwhelmed and the need to withdraw from the child was experienced by families with children diagnosed with PANS/PANDAS as well as other chronic illnesses (Coffey, 2006). However, caregivers of PANS/PANDAS are distinct in their need to research possible causes and treatments without the aid of a medical professional. PANS/PANDAS families were found to face difficulties with the medical community similar to other chronic illnesses, but such difficulties were often the result of the unknown and controversial nature of PANS/PANDAS, which is not the primary obstacle for other chronic illnesses. Frankovich et al. (2018) and Farmer et al. (2018) noted that caregiver burden was higher for parents of children who had to switch schools, missed school frequently, and for those who had to reduce their work hours. The current study followed this pattern but discovered that the caregiver burden was partially due to the reconfiguration of daily life as the parent becomes stay-at-home and the repercussions of losing a part of the household income.

Though PANS/PANDAS is not considered a terminal diagnosis, which may be the case for asthma or cancer, children with PANS/PANDAS may not be able to reach a point of full recovery. Just as it is difficult to see an end to PANS/PANDAS, many families are not able to receive justification for why this occurred in their child. As families reflect on their journey, it is not uncommon to recall early signs but these cannot be confirmed as a factor in the child's ultimate diagnosis. For children with other, more well-known chronic illnesses, parents may find comfort in the ability to navigate treatment with less unknowns (Coffey, 2006). This is

unfortunately not the case for families of children with PANS/PANDAS as treatment protocols vary from child to child; there is no one cure for all. Uncertainty explicitly in regards to symptoms, treatment, and prospective future, was not mentioned in the literature of other chronic illnesses.

As discussed above, PANS/PANDAS is variable in that it is not the result of one clear trigger, nor does it affect families equally. Families of children with PANS/PANDAS suffer from large financial burdens as doctors, treatments, and procedures are not typically covered through insurance. Mettica (2018) detailed that (1) greater symptom severity in children with PANS/PANDAS resulted in greater impact on the family, (2) marriage was a buffer to parental strain, and (3) low family income (less than \$50,000) contributed to a greater impact on the family. The current study goes further in describing some possible reasons why these patterns exist. Greater symptom severity was found to contribute to greater disruption to the parent-child and sibling relationships. In addition, greater symptom severity was explained as requiring more changes to the routines and daily life of PANS/PANDAS families. It was often the more severe cases of PANS/PANDAS that involved the child being removed from school and a parent having to give up their job to provide care. Additionally, more severe symptom presentation often entailed greater strain on the family's resources as more doctor appointments and treatments were necessary. Thus, when family income is lower, families were at a greater disadvantage in their ability to provide full medical attention. This in turn increased parental distress, placing strain on the marriage. For these reasons, the current study found that being married was associated with a decrease in parental strain only if the marriage was positive, stable, and both parents were in agreement on the acceptance of PANS/PANDAS diagnosis, child's severity and treatment plan.

The difference between other childhood chronic illnesses and PANS/PANDAS is highlighted in the way that PANS/PANDAS families describe their experience. The use of trauma metaphors supports the notion that the ability to maintain family functioning is not solely a factor of resilience or preparedness. The unpredictability in family emotional well-being indicates a level of suffering that is divergent from the literature on family functioning as impacted by other childhood chronic illness.

Limitations

As in other studies regarding families and their experience with a childhood chronic illness, some limitations must be acknowledged. First, participants in the online parent forum and interviews may have been over-representing families with more compromised children. Similarly, it is understood that there may be differences between those who willingly participate in studies and those who do not. These families may represent a sub-sample of families who are more likely to be advocates for PANS/PANDAS, use technology and networks, and create strategies to research their child's condition. In addition, those not on Facebook or not in the Facebook groups in which the flier for participation in the interview phase was posted, were less likely to hear about the study. Those unable to devote the time to an in-person interview were also excluded from the study. As mothers are more often the primary caretaker for a child with PANS/PANDAS, it was the mother's perspective that comprised much of the PANS/PANDAS narratives. With this in mind, it would be beneficial to seek more father participation in studies to compare narratives. In addition, the current study did not incorporate teacher accounts which may be useful in future research for understanding how the child's symptoms manifest at school and at home.

Second, it is recognized that the interview study took place in the Northeast of the United States but the PANS/PANDAS population is world-wide. In the online parent forum, most families appeared to be widely distributed across the U.S. Thus, the discussion of difficulties experienced by families regarding schools and the medical community are specific to these regions. Differences in family functioning may be present across countries due to varying healthcare systems, schools, and economic inequalities.

Third, as described in the methodology, the current study reduced the sample from 20 families to eight for the purpose of excluding cases from analysis in which a child was given a previous autism spectrum disorder (ASD) diagnosis. This was done in order to analyze the impact of PANS/PANDAS in the absence of additional stressors that can accompany the navigation of the healthcare system and support services for ASD. However, this reduction led to a predominantly middle class as well as all white/Caucasian sample. As aforementioned, families of children with PANS/PANDAS are considered a hard-to-reach population. Thus, this reduced interview sample provides a view of families who were able to receive a diagnosis and treatment for their child. Families who do not have an official diagnosis but have completed research on their own regarding their child's behaviors and PANS/PANDAS are still accessible through the recruitment procedures completed in this study. The truly inaccessible families are those who have an affected child but no one has identified PANS/PANDAS as a possible diagnosis, neither the healthcare practitioners nor the family. In these cases, families are less likely to be aware of PANS/PANDAS and the resulting networks, such as the Facebook group in which participant recruitment was conducted. We cannot comment on the types of families that are missing from the data but are aware that there are children who are not accessible through doctors nor through the online parent forums or Facebook groups. For these reasons, it is recognized that the

experiences of those who are inaccessible may be different than the ones described in the current study.

Lastly, the current study is a snapshot in time that provides information as it currently stands. PANS/PANDAS is still far from being universally recognized by medical professionals but recognition has improved in the last two decades. Recognition is continuing to change which will likely affect some of the determinants of family functioning. For example, families are often preoccupied with the financial burden that comes with a PANS/PANDAS diagnosis. In the event that insurance companies begin to cover providers and the full range of treatment options, this burden may be lessened or become nonexistent. In a similar fashion, as schools and communities become familiar with PANS/PANDAS, new supports may come to exist, providing a buffer for families. It is the hope that such changes will be made and families will have less disruption to their family functioning.

Implications

As evidenced by the current study, families must adapt to the unexpected and misunderstood nature of PANS/PANDAS while maintaining a level of functionality. However, many families are unprepared or inexperienced in caring for a child with needs outside typical development. Parents must also grapple with the idea that their child will not follow a typical trajectory. This often requires changing parenting tactics, parenting style, and expectations of the child and their future. When children are particularly young, the adaptation to diagnosis can be that much harder as children will require more help in medication administration, regulation of behaviors, and changes to diet or activity. Similarly, the symptomology of PANS/PANDAS may cause family upset if they are particularly life altering, life threatening, or otherwise alarming.

For these reasons, there is a great need for better supports to be in place for these families. Most important is change to the institutional barriers presented earlier.

Families in the interviews and online parent forum noted that if the doctor appointments and treatments were more affordable, their ability to manage the needs of their child would change dramatically. It was found that the lack of answers combined with little awareness was crippling to the families studied. The PANS/PANDAS community needs greater medical acceptance so as to decrease the time lag between onset of symptoms and proper diagnosis. It would be beneficial for pediatricians, specifically, to become well-versed in the signs and symptoms that are indicative of PANS/PANDAS in a child. While pediatricians may not be able to treat the symptoms on their own, acknowledging and understanding parent concerns greatly decreases stress on the family and time spent suffering. In a similar way, providing school nurses and teachers with accurate information to spot signs of PANS/PANDAS in the classrooms could decrease the amount of time spent without diagnosis and treatment. With better understanding and acceptance, schools will be able to provide appropriate accommodations for a child when necessary. Parents and teachers need to be able to have open communication surrounding the child's behavior and capabilities as an indicator of treatment progression or failure.

The experience of families with a child diagnosed with PANS/PANDAS can be understood as including, shifts in close relationships (including changes to the parent-child relationship), adjustment to routines and daily life, and the overall emotional well-being of the family unit, each of which may vary according to the child's symptoms, clinical course, and institutional barriers. As more research is needed to further determine how well the themes and experiences described here generalize to all families in the PANS/PANDAS community, we can begin to better understand the reality of these families. It should be noted that not every family

suffers from these impacts on their support system and resources. With this in mind, if a family were financially stable and had a positive support network (friends/family, child's school, and doctors), they were found to be better equipped to deal with their child's chronic illness. It was these factors, along with the child's symptom severity and family emotional well-being that ultimately made the difference between management of PANS/PANDAS and sustained family functioning, or not. As one parent in the forum explained, "PANDAS has brought us very far from the life we imagined that all of us would be enjoying." Given the current rise in childhood epidemics including newer diagnoses like PANS/PANDAS, understanding the impact on family functioning and well-being is critical for improving outcomes of affected children and the family system.

References

- Atkinson, R., & Flint, J. (2001). Accessing hidden and hard-to-reach populations: Snowball research strategies. *Social Research Update*, 33(1), 1-4.
- Attard, A., & Coulson, N. S. (2012). A thematic analysis of patient communication in Parkinson's disease online support group discussion forums. *Computer in Human Behavior*, 28, 500-506. doi:10.1016/j.chb.2011.10.022.
- Bock K., Stauth C., & Fink, K. (2008). *Healing the new childhood epidemics: Autism, ADHD, asthma, and allergies – the groundbreaking program for the 4-A disorders*. Ballantine Books.
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3(2), 77-101. doi: 10.1191/1478088706qp063oa.
- Braun, V., & Clarke, V. (2012). Thematic analysis. In H. Cooper, P. M. Camic, D. L. Long, A. T. Panter, D. Rindskopf, & K. J. Sher (Eds.), *APA handbook of research methods in psychology* (pp. 57-71). American Psychological Association. doi: 10.1037/13620-004.
- Bury, M. (2001). Illness narratives: Fact or fiction?. *Sociology of Health & Illness*, 23(3), 263-285.
- Chang, K., Frankovich, J., Cooperstock, M., Cunningham, M. W., Latimer, M. E., Murphy, T. K., Pasternack, M., Thienemann, M., Williams, K., Walter, J., & Swedo, S. E. (2015). Clinical evaluation of youth with Pediatric Acute-Onset Neuropsychiatric Syndrome (PANS): Recommendations from the 2013 PANS Consensus Conference. *Journal of Child and Adolescent Psychopharmacology*, 25(1), 3-13. doi: 10.1089/cap.2014.0084.

- Charmaz, K. (2001). Grounded theory. In R. M. Emerson (Ed.), *Contemporary field research: Perspectives and formulations* (pp. 335-352). Waveland Press.
- Charmaz, K. (2006). Coding in grounded theory practice. In K. Charmaz (Ed.), *Constructing grounded theory a practical guide through qualitative analysis* (pp. 42-71). Sage Publications.
- Coffey, J. S. (2006). Parenting a child with chronic illness: A metasynthesis. *Pediatric Nursing, 32*(1), 1-10.
- Doron, H., Hen, M., & Sharabi-Nov, A. (2018). Relationship quality among chronically ill children and their parents. *Journal of Child and Family Studies, 27*, 3866-3876. doi: 10.1007/s10826-018-1228-8.
- Dyson, L., & Fewell, R. R. (1986). Stress and adaptation in parents of young handicapped and nonhandicapped children: A comparative study. *Journal of the Division for Early Childhood, 10*(1), 25-35.
- Emerson, R. M. (2001). The face of contemporary ethnography. In R. M. Emerson (Ed.), *Contemporary field research: Perspectives and formulations* (pp. 27-53). Waveland Press.
- Farmer, C., Thienemann, M., Leibold, C. M., Kamalani, G., Sauls, B., & Frankovich, J. (2018). Psychometric evaluation of the caregiver burden inventory in children and adolescents with PANS. *Journal of Pediatric Psychology, 43*(7), 749-757. doi: 10.1093/jpepsy/jsy014.
- Frankovich, J., Thienemann, M., Pearlstein, J., Crable, A., Brown, K., & Chang, K. (2015). Multidisciplinary clinic dedicated to treating youth with pediatric acute-onset neuropsychiatric syndrome: Presenting characteristics of the first 47

- consecutive patients. *Journal of Child and Adolescent Psychopharmacology*, 25(1), 38-47. doi: 10.1089/cap.2014.0081.
- Frankovich, J., Leibold, C. M., Farmer, C., Sainani, K., Kamalani, G., Farhadian, B., Willett, T., Park, J. M., Sidell, D., Ahmed, S., & Thienemann, M. (2018). The burden of caring for a child or adolescent with pediatric acute-onset neuropsychiatric syndrome (PANS): An observational longitudinal study. *Journal of Clinical Psychiatry*, 80(1), 2-6. doi: 10.4088/JCP.17m12091.
- Gannoni, A. F., & Shute, R. H. (2010). Parental and child perspectives on adaptation to childhood chronic illness: A qualitative study. *Clinical Child Psychology and Psychiatry*, 15(1), 39-53. doi: 10.1177/1359104509338432.
- Gilbert, D. L., & Kurlan, R. (2009). PANDAS: Horse or zebra? *Neurology*, 73, 1252-1253. doi: 10.1212/WNL.0b013e3181beed27.
- Guest, G., Bunce, A., & Johnson, L. (2006). How many interviews are enough?: An experiment with data saturation and variability. *Field Methods*, 18(1), 59-82. doi: 10.1177/1525822X05279903.
- Hennink, M. M., Kaiser, B. N., & Marconi, V. C. (2017). Code saturation versus meaning saturation: How many interviews are enough?. *Qualitative Health Research*, 27(4), 591-608. doi: 10.1177/1049732316665344.
- Herzer, M., Godiwala, N., Hommel, K. A., Driscoll, K., Mitchell, M., Crosby, L. E., Piazza-Waggoner, C., Zeller, M. H., & Modi, A. C. (2010). Family functioning in the context of pediatric chronic conditions. *Journal of Developmental and Behavioral Pediatrics*, 31(1), 1-14. doi:10.1097/DBP.0b013e3181c7226b.

- Jowett, A. (2015). A case for using online discussion forums in critical psychological research. *Qualitative Research in Psychology*, 12(3), 287-297. doi: 10.1080/14780887.2015.1008906.
- Keller, D., & Honig, A. S. (2004). Maternal and paternal stress in families with school-aged children with disabilities. *American Journal of Orthopsychiatry*, 74(3), 337-348. doi: 10.1037/0002-9432.74.3.337.
- Kleinman, A. (1988). *The Illness Narratives: Suffering, Healing, and the Human Condition*. Basic Books.
- Knepper, K. N. K., & Arrington, M. I. (2018). Parents' narratives in an online PHPV forum: Toward a typology of caregiver illness narratives. *Illness, Crisis & Loss*, 26(4), 316-329. doi: 10.1177/1054137316667594.
- Kraybill, O. G. (2019, June 28). *What parents should know about infections and mental health*. PsychologyToday. <https://www.psychologytoday.com/us/blog/expressive-trauma-integration/201906/what-parents-should-know-about-infections-and-mental>.
- Kurlan, R. (1998). Tourette's syndrome and 'PANDAS': Will the relation bear out? *Neurology*, 50(6), 1530-1534. doi: 10.1212/WNL.50.6.1530.
- Kurlan, R., & Kaplan, E. L. (2004). The pediatric autoimmune neuropsychiatric disorders associated with streptococcal infection (PANDAS) etiology for tics and obsessive-compulsive symptoms: Hypothesis or entity? Practical considerations for the clinician. *Pediatrics*, 113(4), 883-886. doi: 10.1542/peds.113.4.883.
- Melerine, C. & Ledet, L. M. (2019, August). PANDAS: What nurses need to know. *Nursing*, 49(8), 46-49. doi: 10.1097/01.NURSE.0000559921.99814.23.

- Mettica, M. (2018). *Family impacts reported by parents raising children with pediatric acute-onset neuropsychiatric syndrome (PANS)* [Unpublished doctoral dissertation]. Texas Woman's University.
- McClellan, C. B., & Cohen, L. L. (2007). Family functioning in children with chronic illness compared with healthy controls: A critical review. *Journal of Pediatrics*, *150*(3), 221-223. doi: 10.1016/j.jpeds.2006.11.063.
- Murphy, T. K., Storch, E. A., Lewin, A. B., Edge, P. J., & Goodman, W. K. (2012). Clinical factors associated with PANDAS. *Journal of Pediatrics*, *160*(2), 314-319. doi: 10.1016/j.jpeds.2011.07.012.
- Murphy, T. K., Patel, P. D., McGuire, J. F., Kennel, A., Mutch, P. J., Parker-Athill, C., Hanks, C. E., Lewin, A. B., Storch, E. A., Toufexis, M. D., Dadlani, G. H., & Rodriguez, C. A. (2015). Characterization of the pediatric acute-onset neuropsychiatric syndrome phenotype. *Journal of Child and Adolescent Psychopharmacology*, *25*(1), 14-25. doi: 10.1089/cap.2014.0062.
- Murthy, D. (2008). Digital ethnography: An examination of the use of new technologies for social research. *Sociology*, *42*(5), 837-855. doi: 10.1177/0038038508094565
- National Institute of Mental Health. (2019). *PANDAS – questions and answers*. <https://www.nimh.nih.gov/health/publications/pandas/index.shtml>.
- Patton, M. Q. (2002). *Qualitative research and evaluation methods* (3rd ed.). Sage Publications.
- Perro, M., & Adams, V. (2017). *What's making our children sick?: How industrial food is causing an epidemic of chronic illness, and what parents (and doctors) can do about it*. Green Publishing.

- Popp, J. M., Robinson, J. L., Britner, P. A., & Blank, T. O. (2014). Parent adaptation and family functioning in relation to narratives of children with chronic illness. *Journal of Pediatric Nursing, 29*, 58-64. doi: 10.1016/j.pedn.2013.07.004
- Riessman, C. K. (2005). Narrative Analysis. *Narrative, Memory & Everyday Life*, 1-7.
- Singer, M. (2013). Studying Hidden and Hard-to-Reach Populations. In Shenshul, J. J. & LeCompte, M. D (Eds.), *Specialized ethnographic methods: A mixed methods approach* (pp. 255-317). AltaMira Press.
- Spradley, J. P. (1979). *Interview: The ethnographic interview*. Holt, Rhinehart & Winston.
- Swedo, S. E., Leonard, H. L., Garvey, M., Mittleman, B., Allen, A. J., Perlmutter, S., Lougee, L., Dow, S., Zamkoff, J., & Dubbert, B. K. (1998). Pediatric autoimmune neuropsychiatric disorders associated with streptococcal infections: Clinical description of the first 50 cases. *American Journal of Psychiatry, 155*(2), 264–271. doi: 10.1176/ajp.155.2.264
- Swedo, S. E., Garvey, M., Snider, L., Hamilton, C., & Leonard, H. L. (2001). The PANDAS subgroup: Recognition and treatment. *CNS Spectrums, 6*(5), 419-426. doi: 10.1017/S1092852900021799.
- Swedo, S. E., Leonard, H. L., & Rapoport, J. L. (2004). The pediatric autoimmune neuropsychiatric disorders associated with streptococcal infection (PANDAS) subgroup: Separating fact from fiction. *Pediatrics, 113*(4), 907-911. doi: 10.1542/peds.113.4.907.
- Swedo, S. E., Leckman, J. F., & Rose, N. R. (2012). From research subgroup to clinical syndrome: Modifying the PANDAS criteria to describe PANS (Pediatric acute-

- onset neuropsychiatric syndrome). *Pediatrics & Therapeutics*, 2(2), 1-8. doi: 10.4172/2161-0665.1000113.
- Swedo, S. E., Seidlitz, J., Kovacevic M., Latimer, M. E., Hommer, R., Lougee, L., & Grant, P. (2015). Clinical presentation of pediatric autoimmune neuropsychiatric disorders associated with streptococcal infections in research and community settings. *Journal of Child and Adolescent Psychopharmacology*, 25(1), 26-30. doi: 10.1089/cap.2014.0073.
- Thomas, E., & Magilvy, J. K. (2011). Qualitative Rigor or Research Validity in Qualitative Research. *Journal for Specialists in Pediatric Nursing*, 16(2), 151-155. doi: 10.1111/j.1744-6155.2011.00283.x.
- Tona, J. T., Bhattacharjya, S., & Calaprice, D. (2017). Impact of PANS and PANDAS exacerbations on occupational performance: A mixed-methods study. *American Journal of Occupational Therapy*, 71(3), 1-9. doi: 10.5014/ajot.2017.022285.
- Weiss, R. S. (1995). Interviewing. In R. S. Weiss (Ed.), *Learning from strangers: The art and method of qualitative interview studies* (pp. 61-199). Free Press.
- Wilbur, C., Bitnun, A., Kronenberg, S., Laxer, R. M., Levy, D. M., Logan, W. J., Shouldice, M., & Yeh, E. A. (2019). PANDAS/PANS in childhood: Controversies and evidence. *Pediatrics & Child Health*, 24(2), 85-91. doi: 10.1093/pch/pxy145.
- Williams, K. A., & Swedo, S. E. (2015). Post-infectious autoimmune disorders: Sydenham's chorea, PANDAS and beyond. *Brain Research*, 1617, 144-154. doi: 10.1016/j.brainres.2014.09.071.

Zamzow, R. (2019, August 25). *How strep throat may spark OCD and anxiety in some kid*. ScienceNews. <https://www.sciencenews.org/article/strep-throat-infections-brain-ocd-anxiety-mental-illness-kids>.

APPENDIX A

Participant Recruitment Flyer

University of Connecticut

Parents/Caregivers Wanted for a Research Study

Families' Experiences with PANS

(Pediatric Acute-Onset Neuropsychiatric Syndrome)

We will be conducting interviews with parents/caregivers of children who have been treated for PANS/PANDAS. The questions will focus on the steps taken to get a diagnosis and treatment, challenges and barriers encountered, impacts of PANS/PANDAS on the family, impacts on children's development and any help received (or not) from schools, healthcare providers, and other support services.

Each family will receive \$75 as a thank you for their time and participation.

The interviews are confidential and information will be provided about procedures for ensuring confidentiality for all study participants. Interviews will take place in locations convenient for families and can be conducted in English or Spanish.

To learn more about this research please contact Dr. Maria LaRusso at 617-470-0321 or maria.larusso@uconn.edu or Dr. César Abadía-Barrero at 617-999-3612 or cesar.abadia@uconn.edu.

APPENDIX B

Institutional Review Board Approval – University of Connecticut



DATE: October 20, 2019

TO: Maria Lorusso,
HDFS

FROM: Pamela I. Erickson, Ph.D.
Chair, Institutional Review Board
FWA# 00007125

RE: Limited IRB Review Protocol #: L19-033, "Families' Experiences with PANS (Pediatric Autoimmune Neuropsychiatric Syndrome)"
Please refer to the Protocol # in all future correspondence with the IRB.
Funding Source: Unfunded
Approved on: October 20, 2019

On October 18, 2019, the Institutional Review Board (IRB) reviewed the above-referenced research study by limited review and determined that modifications were required to secure approval. Those requirements have been met, and the IRB granted approval of the study on October 20, 2019. The IRB determined that study be approved under ~~under~~ 45 CFR 46.104 (d) (2)(iii) - Research that only includes interactions involving educational tests (cognitive, diagnostic, aptitude, achievement), survey procedures, interview procedures, or observation of public behavior (including visual or auditory recording) if the information obtained is recorded by the investigator in such a manner that the identity of the human subjects can readily be ascertained, directly or through identifiers linked to the subjects, and an IRB conducts a limited IRB review to make the determination required by §46.111(a)(7).

Provide an IRB approved information sheet (with electronic validation stamp) to participants in the study.

All investigators at the University of Connecticut are responsible for complying with the attached IRB "Responsibilities of Research Investigators".

Any proposed changes that may affect the limited IRB review status of the research study must be submitted to the IRB for review and approval prior to their implementation.

Attachments:

1. Validated IRB-5 Application and Study Protocol
2. Validated Appendix A
3. Validated Recruitment Material
4. Validated Informed Consent
5. Responsibilities of Research Investigators

Office of the Vice President for Research
Research Compliance Services
355 PASTORAL ROAD, EXTENSION, UNIT 1740
STORRS, CT 06269-0240
Phone: 860-405-3900
Fax: 860-405-3244
compliance@uconn.edu

UConn Institutional Review Board

APPENDIX C

Participant Consent Form

Consent Form for Participation in a Research Study



Principal Investigators: Dr. Maria LaRusso and Dr. César Abadia-Barrero

Study Title: Families' experiences with PANS (Pediatric Acute-Onset Neuropsychiatric Syndrome)

Sponsor: Center for the Study of Culture, Health, and Human Development, University of Connecticut

Overview of the Research

You are being asked to provide consent to participate in a research study. Participation is voluntary. You can say yes or no. If you say yes now you can still change your mind later. Some key points to consider are summarized in this overview, but you should consider all of the information in this document carefully before making your decision.

Research Purpose: This research is being done to better understand family's experiences and journeys of PANS. We want to explore families' understandings of PANS and how the child's symptoms progressed and/or improved over time, as well as the impact of PANS on their child's development and the family's everyday dynamics, social networks, and functioning.

Length: Participation will involve approximately 60-90 minutes of your time.

Procedures: You will be interviewed about your experiences as a parent of a child with PANS, including symptoms, improvements, setbacks, problems with the health care system, and sources of support. During the interview, you do not have to answer any question that you do not want to answer.

Risks: We believe there are no known risks associated with this research study; however, a possible inconvenience may be the time it takes to complete the study.

Benefits: You may not directly benefit from this research; however, we hope that your participation in the study may contribute to knowledge and understanding of families' experiences of PANS, which social supports can use to improve the services that are being offered.

A more detailed description of this research follows.

Introduction

You are invited to participate in a research study to explore family's experiences, understandings and journeys of PANS. We want to understand how families handle the child's symptoms, decide on treatments, and manage the impact of PANS on their child's development and the family's everyday dynamics, social networks, and functioning. You are being asked to participate because you have a child with PANS and have expressed initial interest in participating in the study.

Why is this study being done?

We are conducting this study to better understand how PANS affects families and how families respond to the many challenges that PANS bring to their lives. It is expected that the results of this study will help to inform improvements in programs and social supports available to families with children with PANS.

What are the study procedures? What will I be asked to do?

If you agree to take part in this study, you will be asked to participate in an interview. Interviews are expected to last 60-90 minutes and the questions will focus on your experiences as a parent with a child with PANS, including improvements, setbacks, treatments, and support networks. Interviews will be conducted in available quiet and private settings at your house or other location convenient for you. Interviews will be conducted at times that fit within your schedule. Interviews will be recorded with a digital audio recorder. The interviews will be conducted by the research team (Dr. LaRusso and/or Dr. Abadia-Barrero) who will bring and set up the recorder and then transport the audio file to Dr. LaRusso's research lab to be transcribed. No one else will have access to the recorders or audio files. You might be contacted for follow up questions or clarifications or for future studies.

What are the risks or inconveniences of the study?

We believe there are no known risks associated with this research study; however, a possible inconvenience may be the time it takes to complete the study.

What are the benefits of the study?

You may not directly benefit from this research; however, we hope that your participation in the study may contribute to knowledge and understanding of the challenges that families have in managing PANS, which will inform different program and social supports.

Will I receive payment for participation? Are there costs to participate?

For your participation in this research you will receive \$75. There are no costs to participate in this study.

How will my personal information be protected?

The following procedures will be used to protect the confidentiality of your data. The researchers will keep all study records (including any codes to your data) locked in a secure location. Research records will be labeled with a study code. The code will be derived from a sequential 3 digit number that reflects how many interviews were conducted. A master key that links participants with interview codes will be maintained in a separate and secure location. The master key will be destroyed after 3 years. Digital audio files containing the voices of participants in interviews will be stored in a password protected folder on a computer in the locked research office of Dr. Maria LaRusso at the University of Connecticut. The computer hosting such files will also have password protection to prevent access by unauthorized users. Only the members of the research staff will have access to the passwords. The audio files will be transcribed by a research assistant in Dr. LaRusso's research lab. Transcriptions will be labeled with the study ID and will have no identifying information. The only data that will be shared with others are transcripts with no identifying information and these will be coded with a study ID as described above to help protect your identity. After interviews have been transcribed, audio files will be destroyed. De-identified data will be kept indefinitely. At the conclusion of this study, the researchers may publish their findings. Information will be presented in summary format and you will not be identified in any publications or presentations. Occasionally, illustrative quotes from interview transcripts may be included in publications or presentations and, in such cases, quotes will not contain any information that would reveal the identity of any individual.

We will do our best to protect the confidentiality of the information we gather from you but we cannot guarantee 100% confidentiality. You should also know that the UConn Institutional Review Board (IRB) and Research Compliance Services may inspect study records as part of its auditing program, but these reviews will only focus on the researchers and not on your responses or involvement. The IRB is a group of people who review research studies to protect the rights and welfare of research participants.

Can I stop being in the study and what are my rights?

You do not have to be in this study if you do not want to. If you agree to be in the study, but later change your mind, you may drop out at any time for any reason. There are no penalties or consequences of any kind if you decide that you do not want to participate. During the interview, you do not have to answer any question that you do not want to answer.

Whom do I contact if I have questions about the study?

Take as long as you like before you make a decision. We will be happy to answer any question you have about this study. If you have further questions about this study or if you have a research-related problem, you may contact the principal investigators, *Maria LaRusso at 617-470-0321* or *César Abadía-Barrero at 617-999-3612*. If you have any questions concerning your rights as a research participant, you may contact the University of Connecticut Institutional Review Board (IRB) at 860-486-8802.

Documentation of Consent:

I have read this form and decided that I will participate in the project described above. Its general purposes, the particulars of involvement and possible risks and inconveniences have been explained to my satisfaction. I understand that I can withdraw at any time. My signature also indicates that I have received a copy of this consent form.

Participant Signature:

Print Name:

Date:

Signature of Person
Obtaining Consent

Print Name:

Date:

APPENDIX D

Concept Map of Four Themes Presented in Results

